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A solid pseudopapillary neoplasm of the pancreas in a man presenting with acute pancreatitis: A case report

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ABSTRACT

INTRODUCTION: A solid pseudopapillary neoplasm (SPN) of the pancreas is rare neoplasm that occurs predominantly in young women. The clinical presentation of SPNs is nonspecific, but acute pancreatitis is rare in the reported literature.
PRESENTATION OF CASE: A 36-year-old man was referred to our hospital because of upper abdominal pain and elevation of serum amylase. A computed tomography (CT) scan showed swelling of the pancreas body and a poorly enhanced and indistinct mass in the pancreas body. He was diagnosed with acute pancreatitis. The symptom was improved with conservative treatment, but acute pancreatitis recurred twice during a period of 2 months. Magnetic resonance cholangiopancreatography (MRCP) and endoscopic retrograde cholangiopancreatography (ERCP) showed stenosis of the MPD adjacent to the mass. Distal pancreatectomy was performed because the mass in the pancreas body seemed to cause repeated acute pancreatitis and malignant pancreatic cancer could not be excluded. Immunohistochemically, a diagnosis of SPN of the pancreas was made from the resected specimen.
DISCUSSION: To the best our knowledge, only 6 cases have been reported in the literature concerning the SPN presenting with acute pancreatitis.
CONCLUSION: We report a man with a small SPN of the pancreas presenting with acute pancreatitis and mimicking pancreatic cancer. We should be aware that this rare pancreatic tumor can become a cause of acute pancreatitis.

1. Introduction

A solid pseudopapillary neoplasm (SPN) of the pancreas is rare pancreatic tumor and occurs predominantly in young women [1,2]. The clinical presentation of SPNs is nonspecific, but acute pancreatitis is rare in the reported literature. Herein, we report a man with a small SPN of the pancreas who presented with acute pancreatitis and mimicking pancreatic cancer.

2. Presentation of case

A 36-year-old man was referred to our hospital because of upper abdominal pain and elevation of serum amylase. His medical history was unremarkable except for a duodenal ulcer. He was a moderate drinker but had no history of acute pancreatitis. Laboratory data on admission showed elevated serum levels of pancreatic enzymes: amylase, 1600 IU/L (normal range, 40–129IU/L); *p*-amylase, 1541 IU/L (18–55 IU/L); and lipase, 2243 IU/L (17–57 IU/L). Serum levels of the tumor markers carcinoembryonic antigen and carbohydrate antigen 19-9 were within normal ranges. A contrast-enhanced computed tomography (CT) scan showed diffuse enlargement of the pancreas with inflammatory change in *peri*-pancreatic fatty tissue and a poorly enhanced and indistinct nodule in the pancreas body. The distal side of the main pancreatic duct (MPD) was dilated (Fig. 1**a** and **b**). He was admitted with a diagnosis of acute pancreatitis. The pancreatitis was improved with conservative treatment and he was discharged three weeks later. However, acute pancreatitis recurred twice during a period

Abbreviations: CT, computed tomography; MPD, main pancreatic duct; MRCP, magnetic resonance cholangiopancreatography; ERCP, endoscopic retrograde cholangiopancreatography; Syn, synaptophysin; CgA, chromogranin A; SPN, solid pseudopapillary neoplasm; MRI, magnetic resonance imaging; EUS-FNA, endoscopic ultrasonography guided fine-needle aspiration.

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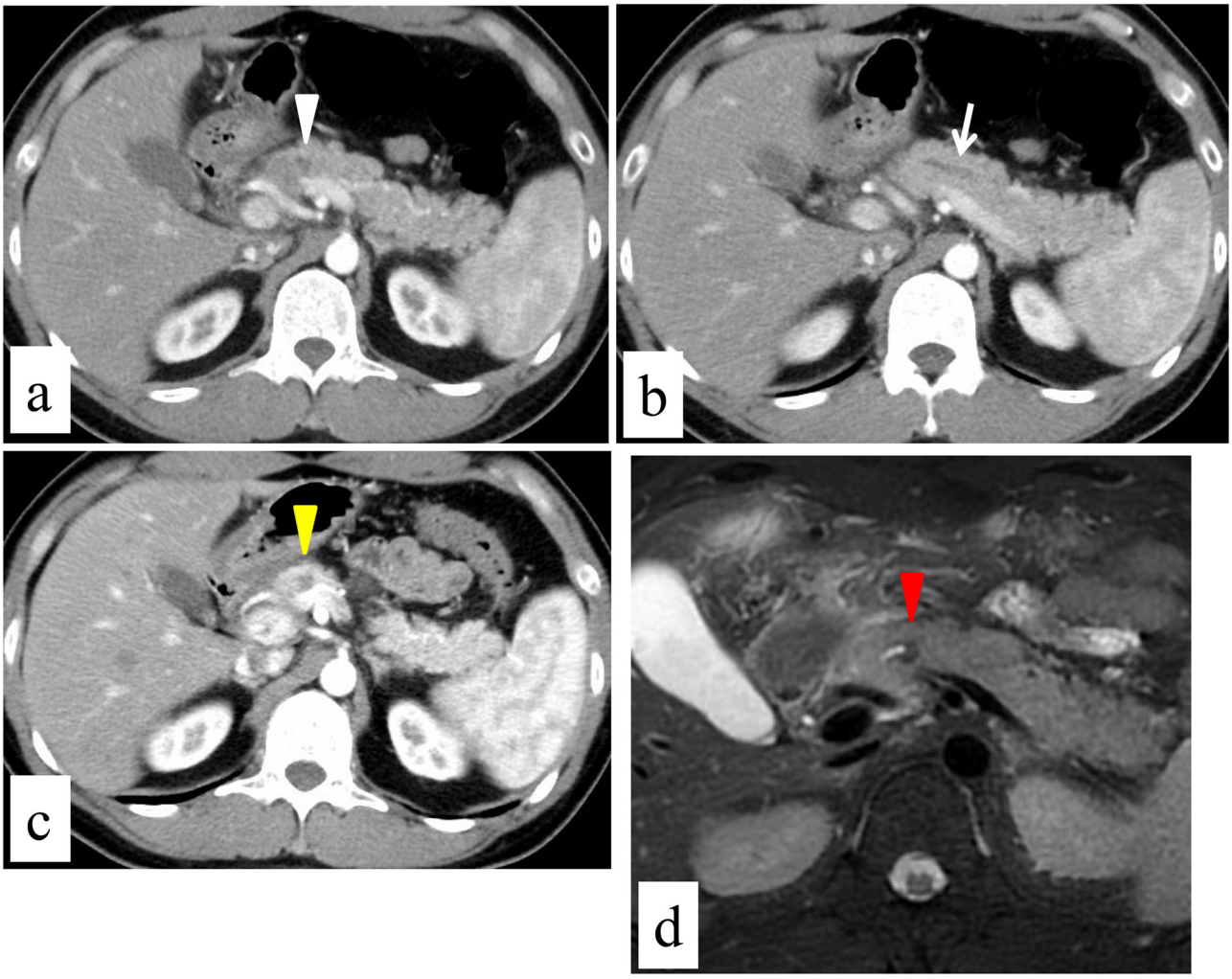

Fig. 1. (**a**) Contrast-enhanced computed tomography (CT) scan showed a poorly enhanced and indistinct nodule in the pancreas body (white arrowhead). (**b**) The distal side of the main pancreatic duct (MPD) was dilated (white arrow). (**c**) The mass in the pancreas body was 10 mm in diameter 53 days later (yellow arrowhead). (**d**) The mass showed low signal intensity on T2-weighted images obtained by magnetic resonance imaging (MRI) (red arrowhead).

of two months with repeated admission and discharge. The mass in the pancreas body gradually became clearer, and a mass of 10 mm in diameter was detected 53 days later by CT (Fig. 1**c**). The mass contained no calcification or cystic lesion. The mass showed low signal intensity on T2-weighted images and diffusion-weighted images obtained by magnetic resonance imaging (MRI) (Fig. 1**d**). Magnetic resonance cholangiopancreatography (MRCP) showed stenosis of the MPD adjacent to the mass (Fig. 2**a**). Endoscopic retrograde cholangiopancreatography (ERCP) also showed stenosis of the MPD (Fig. 2**b**). Cytology of pancreatic juice collected during the ERCP did not reveal malignant cells (Fig. 2**c**). Although a definitive diagnosis could not be made, the recurrent acute pancreatitis was thought to be related to the stenosis of the MPD caused by the mass in the pancreas body. Furthermore, the possibility of a malignant disease such as pancreatic cancer for the mass could not be excluded from the radiological findings. Distal pancreatectomy with regional lymph node dissection was performed. Macroscopically, a well-circumscribed mass, measuring 12 mm, in the pancreas body was observed (Fig. 3**a** and **b**). Microscopically, most of the mass consisted of a fibrous nodule, but an irregularly arranged pseudopapillary structure composed of fairly uniform tumor cells was seen around the fibrous nodule. The tumor cells had proliferated invasively in the pancreatic parenchyma (Fig. 3**c** and **d**). The tumor located adjacent to the MPD and the fibrous and degenerative change had caused a deformity of the wall of the MPD (Fig. 3**e**). The border region between the fibrous nodule and the tumor contained calcification, old hemorrhage and cholesterin crystals (Fig. 3**f**). Immunohistochemically, the tumor was positive for ß-catenin, vimentin, CD10, synaptophysin (Syn) (Fig. 4**a**–**d**), CD56, and neuron-specific enolase (NSE), but negative for chromogranin (CgA) (Fig. 4**e**) and insulin. The Ki67 labeling index was approximately 3% (Fig. 4**f**). Based on these findings, a diagnosis of SPN of the pancreas was made. Lymph node metastasis was not found. The patient's postoperative course was uneventful and he was discharged on the 22nd day after the operation. At a 32-month follow-up after resection, the patient did not have any recurrence of SPN or pancreatitis.

3. Discussion

SPN of the pancreas is a rare neoplasm accounting for only about 0.13–2.7% of all pancreatic tumors [1]. SPN is regarded as a low-grade malignant potential tumor with an excellent prognosis after complete resection. In a large review of the English literature including 718 patients with SPN [2], the male-to-female ratio was 1:9.78 with a median age of 21.97 years and the mean diameter of the tumors was 6.08 cm. The clinical presentation is nonspecific. The majority of patients presented with abdominal