

## Groove pancreatitis vs groove pancreatic adenocarcinoma. Report of two cases and review of the literature

Jeremias Goransky,<sup>1</sup> Fernando A Alvarez,<sup>1</sup> Pedro Picco,<sup>1</sup> Juan C Spina,<sup>2</sup> Martín de Santibañes,<sup>1</sup> Oscar Mazza<sup>1</sup>

<sup>1</sup> Hepato-Pancreato-Biliary Surgery Section, Department of General Surgery.

<sup>2</sup> Department of Clinical Radiology.

Hospital Italiano de Buenos Aires. Ciudad Autónoma de Buenos Aires, Argentina.

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### Summary

Groove pancreatitis (GP) is a rare form of segmental chronic pancreatitis affecting the groove area (anatomic space between the head of the pancreas, the duodenum and the common bile duct). Its clinical and radiological presentation may be similar to groove pancreatic adenocarcinoma (GPA). Nevertheless, treatment and prognosis are totally different. We report two cases of both GP and GPA and review the relevant aspects that may help to clarify the differential diagnosis between these two rare entities. The first patient is a 57-year-old man with a history of chronic alcohol consumption who presented with persistent abdominal pain. The CT-scan findings suggested GP. Due to the persistence of symptoms despite medical treatment, a pancreaticoduodenectomy was performed. Pathologic evaluation confirmed the diagnosis of GP. The second patient is a 72-year-old male who presented with cholestasis and weight loss. The tumor marker CA 19-9 was increased. The CT-scan findings were consistent with duodenal dystrophy. In order to rule out malignancy a pancreaticoduodenectomy was performed. Pathologic evaluation revealed a pancreatic head adenocarcinoma (T3-N1-M0). GP is a rare entity that should be suspected in patients with a history of heavy alcohol consumption who complain of chronic abdominal pain and weight loss. Patients without a clear diagnosis even after a through imaging work-up, or those in whom symptoms are persistent in spite of medical therapy, should undergo surgical exploration.

**Correspondence:** Oscar Mazza

Cabello 3334. CP 1425. Ciudad Autónoma de Buenos Aires, Argentina.

Tel: +54-11 / 4959 0200 Ext 8412, +54-9-11/ 3476 5025

Fax: +54-11/ 4958 2200

E-mail: oscar.mazza@hospitalitaliano.org.ar

**Key words.** Groove pancreatitis, groove pancreatic adenocarcinoma, chronic pancreatitis, paraduodenal pancreatitis.

### Pancreatitis del surco vs adenocarcinoma pancreático del surco. Reporte de dos casos y revisión de la literatura

#### Resumen

La pancreatitis del surco (PS) es un proceso inflamatorio crónico del páncreas poco frecuente que se caracteriza por la afectación segmentaria del espacio anatómico entre la cabeza del páncreas, el duodeno y la vía biliar. Su presentación clínica y radiológica puede ser similar al adenocarcinoma pancreático del surco (APS), siendo su diagnóstico diferencial dificultoso. Sin embargo, su tratamiento y pronóstico son completamente diferentes. En el presente trabajo presentamos 2 casos de PS y APS, y revisamos los aspectos relevantes que pueden ayudar a aclarar el diagnóstico diferencial entre estas dos entidades poco frecuentes. El primer paciente es un hombre de 57 años con antecedentes de consumo crónico de alcohol que consultó por dolor abdominal persistente. Los hallazgos tomográficos fueron sugestivos de PS. Debido a la persistencia de síntomas a pesar del tratamiento médico, se llevó a cabo una duodeno-pancreatectomía cefálica. La anatomía patológica confirmó el diagnóstico de PS. El segundo paciente es un varón de 72 años con colestasis, pérdida de peso y un CA 19-9 elevado. La tomografía demostró distrofia duodenal. Con el fin de descartar malignidad se llevó a cabo una duodenopancreatectomía cefálica. La anatomía patológica reveló un adenocarcinoma pancreático (T3-N1-M0). La PS es una entidad poco frecuente que debe sospecharse en

*pacientes con antecedentes de consumo de alcohol en exceso que se presentan con dolor abdominal crónico y pérdida de peso. Los pacientes sin un diagnóstico claro, incluso después de un minucioso estudio por imágenes, o aquellos en los que los síntomas son persistentes a pesar del tratamiento médico, deben ser sometidos a una exploración quirúrgica.*

**Palabras claves.** *Pancreatitis del surco, adenocarcinoma pancreático del surco, pancreatitis crónica, pancreatitis paraduodenal.*

Groove pancreatitis (GP) is a rare form of segmental chronic pancreatitis affecting the anatomic space between the head of the pancreas, the duodenum and the common bile duct. This type of pancreatitis was first reported by Becker et al in 1973,<sup>1</sup> but was Stolte et al who communicated in 1982 the first series of patients and described the characteristics of this entity.<sup>2</sup> It is also known as paraduodenal pancreatitis, cystic dystrophy of heterotopic pancreas and pancreatic hamartoma of the duodenum. All these denominations refer to its morphological characteristics. Brunner's glands hyperplasia and a fibrous scar in the groove area with duodenal stenosis due to cyst dystrophy or heterotopic pancreas represent the most frequent histological findings.<sup>3,4</sup>

Becker classified groove pancreatitis into a pure form which only affects the groove area and a segmental form in which all the head of the pancreas is affected.<sup>2,5</sup> The pathogenesis seems to be caused either by a complicated heterotopic pancreas, a disturbance on the pancreatic outflow at the Santorini's duct or chronic alcohol consumption.<sup>2-4,6</sup> The reported prevalence varies significantly, with rates ranging between 2.7% and 24.5% in

pancreaticoduodenectomies for chronic pancreatitis.<sup>2,5,7,8</sup> The main challenging aspect of GP is its differential diagnosis from groove pancreatic adenocarcinoma (GPA). GP can be treated by conservative measures. Surgical resection should be reserved for patients with severe symptoms or in order to rule out malignancy.

The aim of this article is to report two cases of GP and GPA and review the relevant aspects that may clarify the differential diagnosis between these two rare entities.

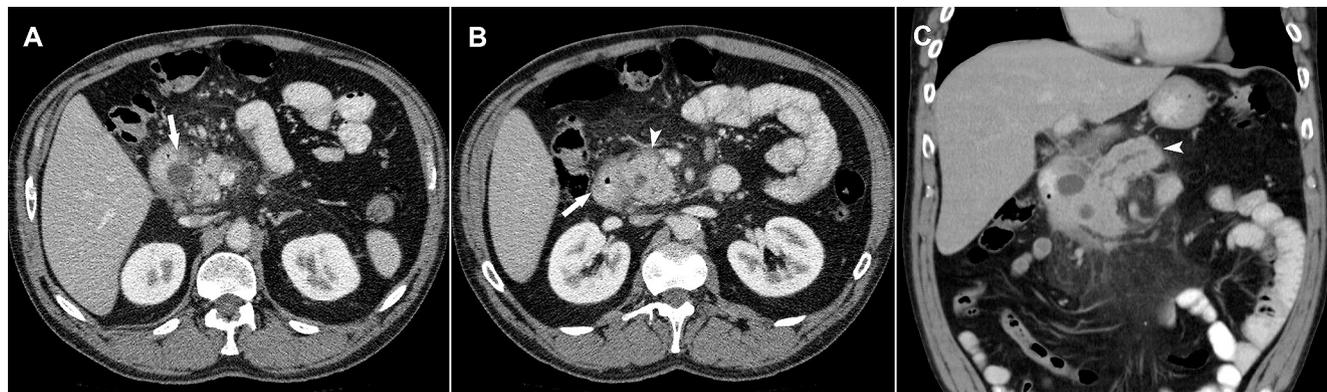
## Case reports

### Case 1

We present a case of a 57-year-old man with a history of chronic alcohol consumption and acute pancreatitis with multiorgan failure eight months before referral to our centre. He presented with persistent epigastric abdominal pain. Laboratory tests and tumor markers were within normal limits. An abdominal CT-scan examination revealed swelling of the pancreatic head and duodenal cyst dystrophy (Figure 1).

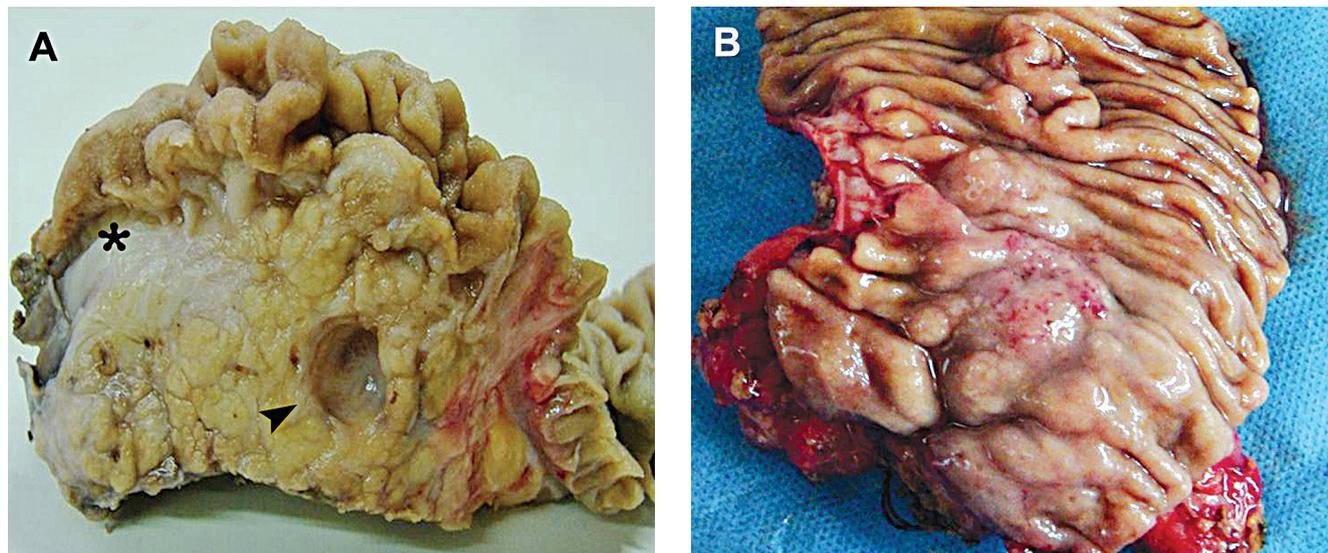
Under the presumptive diagnosis of GP he started a medical treatment. After a 3-month period he did not improve his symptoms and due to the difficulty of ruling out malignancy a pylorus preserving pancreaticoduodenectomy was performed (Figure 2). He developed a type A pancreatic fistula according to the International Study Group of Pancreatic Fistula (ISGPF) classification,<sup>9</sup> and was discharged 8 days after surgery. Microscopic examination revealed Brunner gland hyperplasia and cystic dystrophy in the submucosa of the duodenal wall (Figure 3). After 15-months of follow-up the patient remains asymptomatic.

**Figure 1.** Pre-operative CT-scan of the abdomen shows findings consistent with GP.



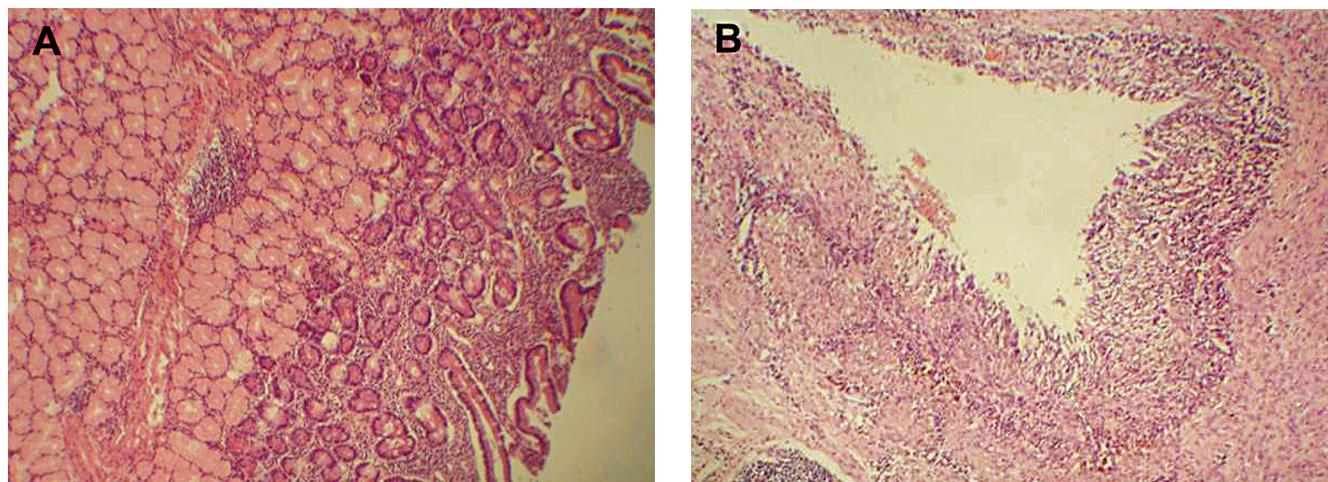
(A) Duodenal wall thickening with luminal narrowing and a paraduodenal cyst (arrow) is evidenced in axial plane images. (B) Swelling of the pancreatic head (arrow head) and stenosis of the duodenal wall (arrow). (C) A dilated main pancreatic duct is seen after coronal reconstruction (arrow head).

**Figure 2.** Gross examination of the specimen after surgical resection.



(A) A whitish fibrous scar (asterisk) with a 1x1 cm long paraduodenal cyst (arrow head) is seen in the groove area as well as a thickened duodenal wall. (B) Duodenal mucosa looks edematous and has a polypoid appearance.

**Figure 3.** Microscopic examination.



(A) Brunner gland hyperplasia. (B) Chronic pancreatitis with mononuclear inflammatory infiltrate in the groove area and cystic dystrophy in the duodenal submucosa is seen.

## Case 2

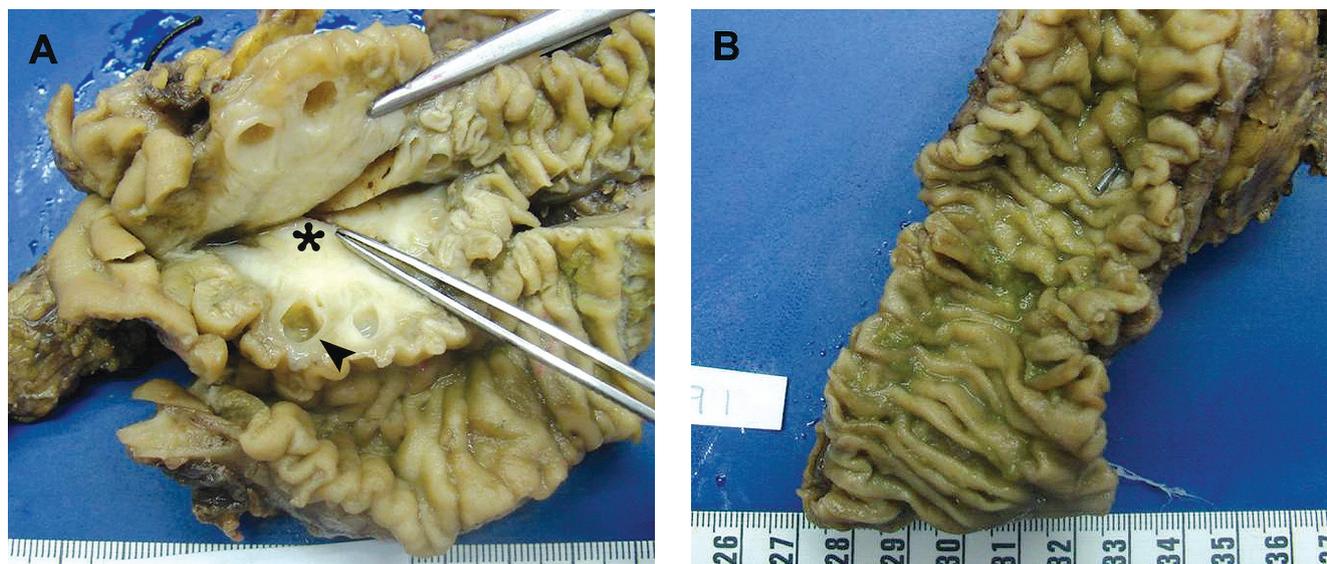
A 72-year-old male with no relevant medical history presented with severe obstructive jaundice and weight loss. The laboratory tests confirmed cholestasis and an elevated CA 19-9 tumor marker (1,470 u/ml). Transabdominal ultrasonography showed biliary tract dilation and a CT-scan evidenced duodenal wall thickening with cyst dystrophy (Figure 4). An endoscopic retrograde cholangiopancreatography (ERCP) demonstrated duodenal narrowing with normal aspect of the mucosa as well as a large stenosis of the common bile duct with marked

biliary dilation. A plastic stent was endoscopically placed. Even though the imaging work-up suggested duodenal dystrophy, a pancreaticoduodenectomy was performed to rule out malignancy (Figure 5). He had a full recovery and was discharged 9 days after surgery.

The microscopic evaluation revealed groove pancreatic adenocarcinoma with infiltration of the duodenal submucosa and tumor-free surgical margins (T3-N1-M0). The patient received adjuvant chemotherapy and remains with no progression of disease after 9 months of follow-up.

**Figure 4.** Pre-operative CT-scan.

(A) Axial plane images showed a hypodense lesion in the groove area with paraduodenal cysts and a dilated biliary tree. (B) Coronal reconstruction showed a thickened duodenum with cysts on its wall (arrow) and dilated biliary ducts (asterisk).

**Figure 5.** Gross examination of the specimen in a patient with a groove pancreatic adenocarcinoma.

(A) A pale and indurated lesion located in the pancreatic head (asterisk) with cyst formation on the duodenal wall (arrow). (B) The duodenal mucosa appears normal.

## Discussion

The two largest series of GP were published by Stolte et al in 1982,<sup>2</sup> and Casati et al in 2009.<sup>7</sup> Despite many contributions over 30 years, the main challenging aspect of GP is still its differentiation from GPA, and there is only scarce literature that compares these two entities in the preoperative scenario.<sup>10,11</sup>

As presented in our first patient, GP appears in men

with history of heavy alcohol use in the fourth and fifth decades of life, in a similar way of chronic pancreatitis. The main symptoms include recurrent abdominal pain, vomiting and weight loss. Jaundice is extremely rare but may be present if late stenosis of the common bile duct occurs.<sup>2,7,12</sup> In contrast, GPA generally appears in the sixth or seventh decades with less incidence of alco-

hol abuse. Main symptoms present in these patients are jaundice and weight loss.<sup>2,7,11</sup> Concerning the laboratory tests, GP and GPA may show a slight elevation of pancreatic enzymes whereas the tumor markers, the bilirubin levels and alkaline phosphatase are rarely elevated in GP.<sup>10,13</sup>

In ultrasound examination GP typically displays a hypoechoic mass in the groove area and the common bile duct may be slightly or not dilated at all. Endoscopic ultrasound may show cyst dystrophy of the duodenal wall with smooth tubular stenosis of the intrapancreatic common bile duct as well as minor irregularities of the main pancreatic duct might be revealed,<sup>14</sup> whereas ultrasonography in GPA usually reveals an irregular infiltrative hypoechoic mass in the pancreatic head. Typically the pancreatic and common bile ducts are dilated with an abrupt and short stenosis of the latter.<sup>10,14</sup>

Currently, most authors perform endoscopic fine needle aspiration as a routinely diagnostic procedure, even though its usefulness is still controversial.<sup>10,15</sup> The role of either CT-scan and magnetic resonance imaging (MRI) has not been established yet.<sup>7,10,16</sup> Generally the CT imaging findings in GP and GPA are overlapped. In both cases the CT-scan reveals a hypodense mass with delayed contrast enhancement as was seen in our patients.<sup>16</sup> However, some specific characteristics such as the duodenal cyst dystrophy and the contrast enhancement might help in the differential diagnosis. GP usually presents a hypodense mass with a patchy enhancement in the portal venous phase, together with duodenal cystic dystrophy. In contrast, a hypodense mass with peripheral enhancement is displayed in GPA and cystic dystrophy of the duodenum is exceptionally seen.<sup>16,17</sup> In MRI, GP and GPA image features appear as hypo-intense on T1 and hypo-iso or slightly hyper-intense on T2-weighted images. Magnetic resonance colangiopancreatography (MRCP) in GP reveals a smooth tubular stenosis of the common biliar duct (CBD) with a normal or slightly dilated main pancreatic duct.<sup>18</sup> In the other hand, as in our second patient, GPA usually presents with abrupt stenosis of the CBD and pancreatic duct dilatation. GP usually reveals stenosis of the duodenal lumen and a swollen polypoid mucosal surface as endoscopic findings. In GPA the endoscopy is usually normal or shows an extrinsic compression.<sup>10,15</sup>

Regarding the treatment options, GPA must undoubtedly be resected in the absence of contraindications. On the contrary, conservative measures including analgesics, pancreatic enzymes and abstinence from alcohol

usually are initially successful in GP.<sup>12,15</sup> However there are few reports in the literature concerning the medical treatment of GP with satisfactory long-term results and consequently most patients end up in surgery because of persistent symptoms or to rule out malignancy.<sup>7,8,12</sup> This led the majority of authors to propose surgery as the best treatment with satisfactory long-term outcomes. The largest experience was presented by Casati et al with 58 cases of GP after 882 pancreaticoduodenectomies for diverse pathologies.<sup>7</sup> This series revealed that 76% of the patients had complete disappearance of pain in a median follow-up of 96.3 months. Other surgical procedures have been attempted, such as endoscopic cyst drainage or organ preserving techniques like pancreaticojejunum derivation.<sup>19,20</sup> However there is not enough evidence of long-term results and the number of patients included was small.

The main characteristics that help the differential diagnoses between both entities are summarized in Table 1.

**Table 1.** Differential diagnoses between the two entities.

	Groove pancreatitis	Groove pancreatic adenocarcinoma
<b>Epidemiology</b>	Age 40 to 50 years old Heavy alcohol abuse	Age 60 to 70 years old
<b>Symptoms</b>	Abdominal pain, vomiting, weight loss	Weight loss, jaundice
<b>Tumor markers, bilirubin, alkaline phosphatase</b>	Usually normal	Increased
<b>Ultrasound</b>	Hypoechoic mass CBD: slightly or not dilated	Irregular infiltrative hypoechoic mass. CBD: dilated
<b>CT-scan</b>	Duodenal cyst dystrophy, hypodense mass with patchy enhancement	Hypodense mass with peripherical enhancement
<b>MRCP</b>	Smooth tubular stenosis of the CBD	Abrupt stenosis of the CBD
<b>Endoscopy (duodenal mucosa)</b>	Edematous and polypoid aspect	Normal
<b>Surgery</b>	To rule out malignancy Persistence of symptoms	Always

CBD: common bile duct, MRCP: magnetic resonance colangiopancreatography.

We conclude that groove pancreatitis should be suspected in patients with a past history of chronic alcohol consumption who complain of chronic abdominal pain and weight loss. The differential diagnosis with head pancreatic adenocarcinoma remains challenging among physicians. When clinical presentation, laboratory tests and imaging work-up are characteristic, the diagnosis of

GP is more likely to be possible if interpreted by experienced surgeons and radiologists as a team effort. When the diagnosis is clear, conservative treatment is the first therapeutic approach. Radical surgery (pancreaticoduodenectomy) should be performed at the presence of persistent symptoms despite medical therapy, or when diagnosis of malignancy cannot be ruled out.

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