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Groove Pancreatitis or Pancreatic Neoplasm: A Surgical Dilemmas

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Abstract **Background:** Groove pancreatitis (GP) is a rare condition characterized by inflammatory fibrosis of the space between the dorsal-cranial part of the head of the pancreas, duodenum and common bile duct. Surgery is often the choice of treatment. **Case report:** A 44-year-old male patient with a history of a high alcoholic intake came with epigastric pain associated with a weight loss. An abdominal ultrasonography, a computer tomography (CT) and a gastro-duodenoscopy showed a mass of the pancreatic head with duodenal infiltration suggesting for adenocarcinoma of the pancreas. An endoscopic ultrasound - fine needle aspiration (EUS-FNA) did not show any sign of chronic inflammation. The patient underwent Whipple procedure. Histologic examination confirmed chronic inflammation in the groove area with a ruptured pancreatic-type cyst and some pseudocysts in depth of the wall. **Conclusion:** The diagnosis of GP is challenging. Radiological and endoscopic findings and symptoms are often not specific and they could also be present in patients with adenocarcinoma of the pancreas. The pancreaticoduodenectomy is often the choice of treatment when symptoms do not improve or when there is uncertainty over the diagnosis.

Keywords: *pancreatic neoplasm, pancreatitis, chronic pancreatitis, groove pancreatitis*

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1. Introduction

GP is a rare type of segmental chronic pancreatitis characterized by fibrous scars that affects the anatomical area between the dorso-cranial pancreatic head, the duodenum and the common bile duct. Most patients with GP are aged 40-50, predominantly male with a history of alcohol abuse. A differential diagnosis of GP from peripancreatic cancer is clinically important. The purpose of this study is to report a case and review the current clinical, radiological and pathological characteristics of GP related to groove pancreatic carcinoma.

2. Case Report

A 44-year-old male patient with a history of high alcoholic intake came to our Emergency Department with epigastric pain present since 6 months, not responding to any analgesic drug and associated with a weight loss of 8 kilograms.

Alanine transaminase, Aspartate transaminase, alkaline phosphatase, bilirubin, amylasaemia and hemogram were within normal range at presentation. An abdominal ultrasonography evidenced a hypoechoic mass of the head of the pancreas. CT confirmed a mass with duodenal infiltration and two little hypodense simil-cystic areas in the lesion, suggesting adenocarcinoma of the pancreas (Figure 1). A duodenal wall thickening with three 1 cm cysts was shown during an abdominal EUS. Peripancreatic fluid and dense strands were also seen. The pancreatic hypoechoic mass with not well defined margins, determining neither Wirsung nor common bile duct dilation, was assessed to be probably inflammatory. EUS-FNA did not show any neoplastic cell. A gastro-duodenoscopy evidenced a 6 cm lesion in the second part of the duodenum involving the papilla, but histologic examination of the biopsies found only signs of chronic inflammation. Tumor markers, carcinoembryonic antigen and carbohydrate antigen 19-9, were also negative. The Patient underwent Whipple procedure (Figure 2), with no complications and was discharged on the 14th postoperative day.

Histologic examination on the resected specimen demonstrated Brunner gland hyperplasia (Figure 3*a*), enormous thickening of muscularis mucosa (see for comparison the normal muscolaris propria at the edge of the lesion, Figure 3*c*, compared with the thickened one in Figure 3*d*) with intense and diffuse chronic inflammatory infiltrate, with distortion of muscular fibers (Figure 3*b*) with involvement of the groove area. In the depth of the distorted muscularis propria there was a partially

preserved pancreatic-type cyst (Figure 3*e*), that reacted with anti-CK19 antibody (Figure 3*f*), some pseudocysts with inspissated amorphous eosinophilic material (Figure 3*g*). These findings appeared consistent with the diagnosis of pancreatitis confined to the "groove area". One year after the operation the patient is completely asymptomatic without recurrence of pancreatitis. Evidence of exocrine insufficiency and significant weight loss are absent.

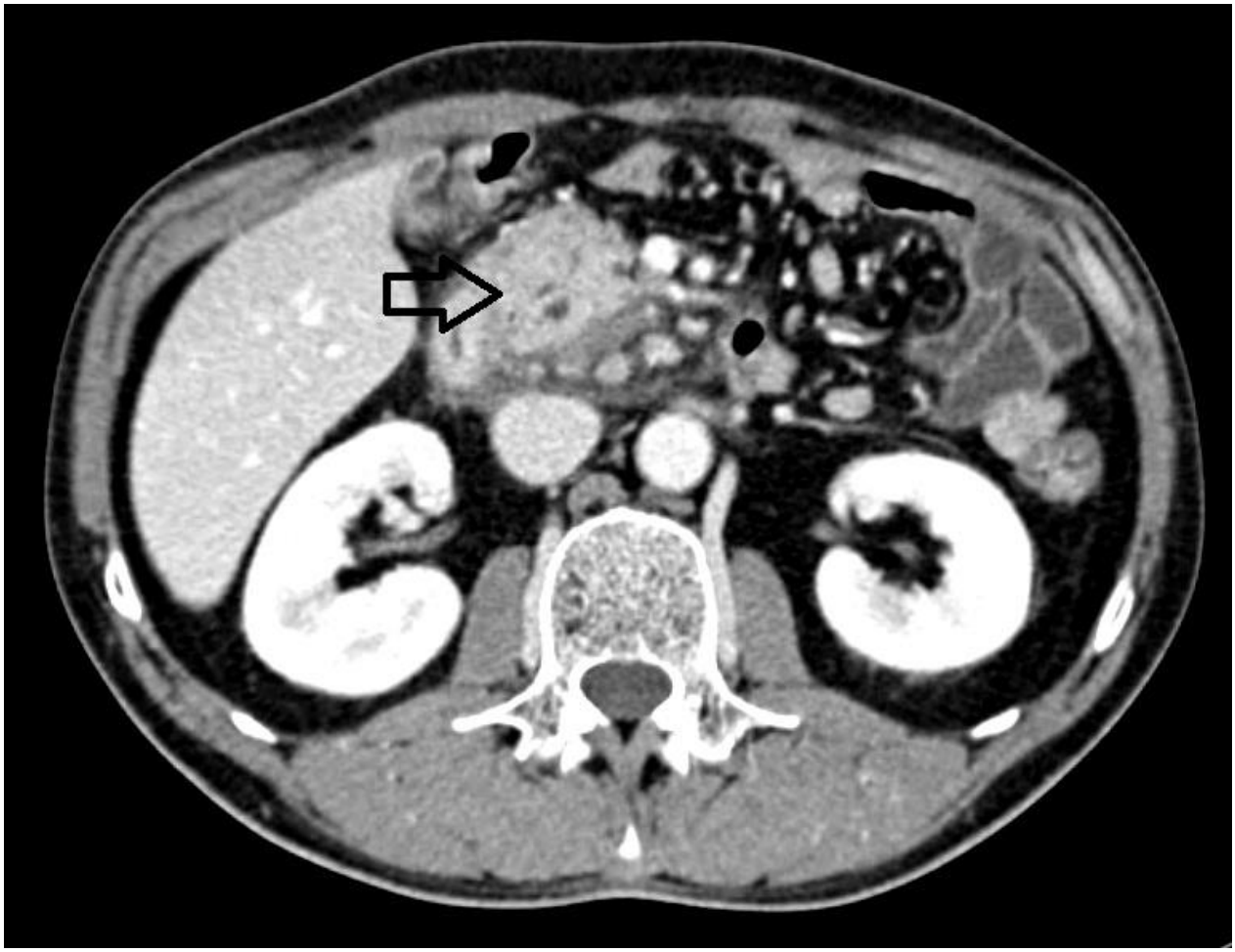

Figure 1. In the arterial phase, the CT revealed swelling of the pancreatic head, 5 cm hypodense mass (black arrow) with multiple cystic structures, and partial duodenal wall thickening with luminal narrowing

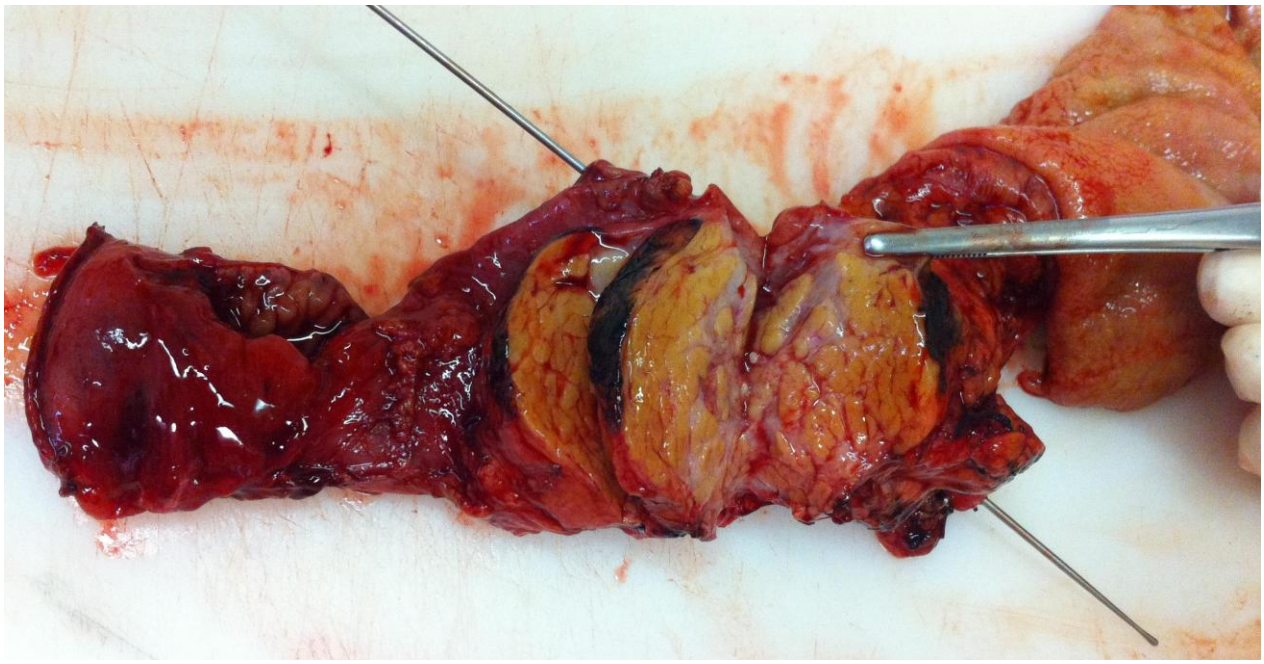

Figure 2. The resected specimen of the duodenum and the head of pancreas. The cut hand section shows the characteristic feature of pancreatic parenchyma in GP. A forceps was introduced into the intrapancreatic biliary tract

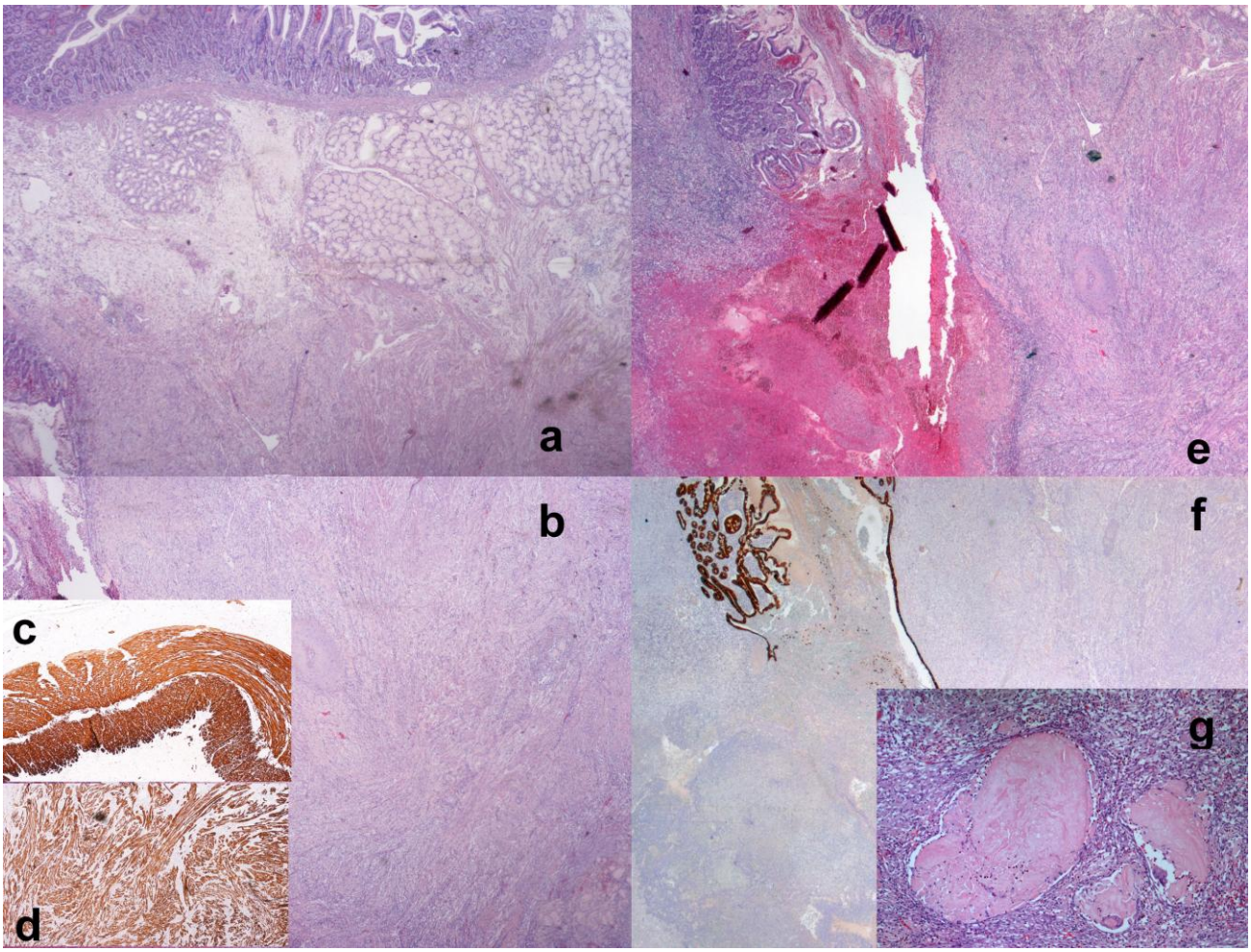

Figure 3. At low magnification, it is evident a Brunner gland hyperplasia (*a*), thickened muscularis propria (*b-c-d*) with a partially preserved epithelial cyst (*e, f*) and some pesudeocysts full of amorphous eosinophilic material (*g*)

(*a* and *b*: Hematoxylin-eosin, original magnification 2.5 X, C-D immunostain for Desmin, original magnification 5X; *e*: Hematoxylin-eosin, original magnification 2X; *f* immunostain for CK19, original magnification 2X; g, Hematoxylin-eosin, original magnification 5X)

3. Discussion

An unusual form of chronic pancreatitis occurring predominantly in and around the duodenal wall has been reported under various names, as cystic dystrophy of heterotopic pancreas [1], pancreatic hamartoma of duodenum [2], para-duodenal wall cyst, myoadenomatosis [3] and GP. [4] This affection, that was described for the first time in the 1970s by Becker et al. as "segmentäre pankreatitis" or "Rinnenpankreatitis" [4], was named "Groove Pancreatitis" by Stolte et al. in 1982. [5] GP was described by Stolte et al. as a special form of segmental pancreatitis characterized by fibrous scars of the anatomic space between the dorso-cranial part of the head of the pancreas, the duodenum and the common bile duct. [5] Adsay and Zamboni published a review in which they unified under "paraduodenal pancreatis" the three concepts of "GP", "cystic dystrophy of heterotopic pancreas" and "paraduodenal wall cyst". [6] Becker [7] and Stolte [5] classified GP into a pure form, in which scarring is localized to the groove area, and a segmental form, in which scarring involves both the groove and the head of the pancreas with stenosis of the pancreatic duct. GP is frequently associated with ectopic pancreatic tissue in the duodenal wall [8].

GP is a rare disease and this is the reason because its incidence is unknown, but it accounts for 19.5-24.4% of pancreaticoduodenectomies performed to treat symptomatic chronic pancreatitis. [5,7] GP presents more frequently in males, in their fourth and fifth decade, with moderate or severe alcohol history [5,7].

The pathogenesis of GP is still unknown. Anatomical or functional obstruction of the minor papilla is thought to be the most credible explanation. The viscosity of the pancreatic juice changes following alcohol consumption and smoking. Pancreatitis in the groove area might arise due to outflow obstruction of the pancreatic juice.[9] Several other factors may be important in pathogenesis, as peptic ulcers, history of gastrectomy, biliary disease, true duodenal-wall cysts, and pancreatic heterotopia in the duodenal wall. [5,10,11,12] Stasis of the pancreatic juice in the dorsal pancreas caused by Brunner gland hyperplasia, could lead to pancreatitis in the groove area. Brunner gland hyperplasia is considered to arise from an increase in cholecystokinin-pancreozymin or gastrin levels [5].

Most common clinical symptoms are weight loss upper postprandial abdominal pain with postprandial nausea and vomiting due to duodenal stenosis caused by inflammatory changes. Symptoms can vary in duration. [8,13] Unlike adenocarcinoma of the head of the pancreas or distal common bile duct, obstructive jaundice is uncommon in GP. The commonest clinical complications are gastrointestinal

hemorrhage, perforation, chronic debilitating abdominal pain, recurrent pancreatitis, duodenal stenosis and risk of malignant transformation of ectopic pancreas [5,8].

Blood tests often show elevation of serum hepatic enzymes [11,14], whereas tumor markers are rarely elevated [15].

Abdominal ultrasound usually shows a hypoechoic mass with distinct appearances reflecting the ongoing pathologic processes in the course of the disease. The main feature of early GP observed is hypoechoic bandlike area in the "groove area" associated with a moderate thickening of the second portion of the duodenum and heterogeneity of the dorso-cranial part of the pancreatic head. [16] In late GP, the sonographic findings usually seen are a hyperechoic thick duodenal wall and a hyperechoic pancreatic head with anechoic ductal structures due to pathognomonic myoadenomatoid proliferation and fibrosis of the adjacent pancreas. In the latest stages of GP, the hypoechoic area may be absent [17].

CT scan often reveals a hypodense mass between the pancreatic head and duodenal wall [11].

Magnetic resonance (MR) imaging usually shows a sheet-like hypointense mass on T1-weighed images, and isointense or slightly hyperintense on T2-weighed images, with delayed contrast enhancement. [10,18] Cysts in the duodenal wall can be shown on T2-weighed images [11].

Endoscopic retrograde cholangiopancreatography (ERCP) can demonstrate distal smooth tubular stenosis of the common bile duct, or rarely, irregularity and dilatation of the Santorini duct and its branches, sometimes with intraductal stones. Biopsy through the duodenum during ERCP is also useful for diagnosis. The pancreatic biopsy in GP shows evidence of chronic pancreatitis with extensive fibrosis, acinar involution and intimal fibrosis of the pancreatic arterioles. [13,19] Upper gastrointestinal endoscopy often reveals inflamed duodenal mucosa and stenosis of the descending part of the duodenal lumen. [20,21] Endoscopic ultrasonography (EUS) provides an accurate location and extension of the disease. [22] This diagnostic tool usually reveals irregular hypoechoic mass in common bile duct due to disease penetration [23].

Characteristic pathological findings in GP are cystic lesions in the duodenal wall, Brunner's gland hyperplasia, dilation of Santorini's duct, fibrosis in the adjacent pancreas and soft tissue and protein plaques in the pancreatic duct. Microscopy reveals thickening of the duodenal submucosal, the muscle layers and the adjacent pancreatic tissue caused by extensive fibrosis [5,12].

This rare but distinct entity is often confused clinically with ampullary neoplasms, duodenal tumours, duodenal hamartoma, cystic tumours of the head of the pancreas and acute or autoimmune pancreatitis [12,24].

The distinction between GP and pancreatic head adenocarcinoma is often difficult on imaging. [11,16,23] Differential diagnosis could be easily reach through histological findings. EUS and transduodenal biopsy during ERCP are useful diagnostic tool to distinguish GP from pancreatic cancer. [21,23] Smooth, long and regular stenosis of the common bile duct is seen in GP while abrupt and irregular ductal stenosis with obstructive jaundice is usually seen in pancreatic adenocarcinoma. Cystic lesions are more often present in the duodenal wall in GP than in pancreatic carcinoma. [7] Gabata et al. reported nine cases of histologically proven carcinoma of the head of the pancreas found in the groove area whose imaging findings resembled those of GP. [16] A high index of suspicion is needed to detect this condition called "pancreatic groove carcinoma".

Conservative treatment including analgesics, recovery of pancreatic function, and abstinence from alcohol/smoke are usually useful to treat initial symptoms. However, these treatments may be temporarily successful. [25] In literature, endoscopic stenting of a stenotic or occluded minor duct has been reported with good initial results, but the long-term clinical course remains obscure [26].

Surgery approach is inevitable when symptoms do not improve or when there is uncertainty over the diagnosis with pancreatic carcinoma. The most common surgical procedures are the pylorus-preserving pancreaticoduodenectomy and the Whipple procedure. [27] In a study by Casetti et al [28], complete pain relief was reported in 76% of the patients undergone surgical procedure, whereas remainders have occasionally relapsing abdominal pain. Egorov et al [29] reported a new surgical approach carried out on two patients who were successfully treated by two modifications of a pancreas-preserving duodenal resection with reimplantation of the bile and pancreatic ducts into the neoduodenum. The authors have claimed that this technique may be an alternative to the Whipple procedure in case of mild changes of the orthotopic gland.

The mean follow-up of patients affected by GP varies greatly and the longest one reported by Casetti et al [28] is about 8 years.

4. Conclusion

GP is a rare disease whose pathogenesis remains controversial. Patients affected by GP present various clinical features, such as biliary obstruction, duodenal stenosis and pancreatic mass. The clinic-pathologic and radiologic features of GP could often masquerade a pancreatic carcinoma. The diagnosis is often difficult and it can lead to surgical dilemmas. In controversial cases, pancreatic biopsy can help to reach diagnosis. GP can be treated by conservative medical measures, but surgery is often required because of the severity of the clinical symptoms and in order to rule out malignancy when it is strongly suspected. Despite several cases have been reported in literature, this disease still remains underdiagnosed because of the lack of awareness of this condition.

Statement of Competing Interests

The authors have no competing interests

List of Abbreviations

GP: groove pancreatitis
CT: computer tomography
EUS: endoscopic ultrasonography
FNA: fine needle aspiration
ERCP: Endoscopic retrograde cholangiopancreatography
MR: Magnetic resonance.

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