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Groove Pancreatitis: Clinical Cases and Review of the Literature

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Keywords
Groove pancreatitis · Pancreatoduodenectomy · Paraduodenal pancreatitis

Abstract
Introduction: Groove pancreatitis (GP) is a type of chronic segmental pancreatitis that affects the pancreatoduodenal groove area, and it is often misdiagnosed. Outflow obstruction of the minor papilla associated with alcohol consumption seems to be the main pathophysiological mechanism, and it affects mainly middle-aged males. Symptoms include nausea and postprandial vomiting from gastric outlet obstruction, weight loss, and abdominal pain. Despite modern advances, such as radiological and endoscopic methods, distinction between GP and pancreatic cancer remains a challenge, and histological examination is sometimes necessary. When a diagnosis can be obtained without a surgical specimen, management can be conservative in the absence of acute or chronic complications. ***Case Presentation:*** The authors present 2 clinical cases which portray the diagnostic workup and management decisions of this entity. ***Discussion/Conclusion:*** GP is a clinical entity, offering diagnostic and therapeutic challenges. Imaging exams are crucial in the diagnosis and follow-up, but surgery may be necessary in a significant number of cases due to the incapacity to rule out malignancy.

Pancreatite da goteira: Casos clínicos e revisão da literatura

Palavras Chave
Pancreatite da goteira · Pancreatoduodenectomia · Pancreatite paraduodenal

Resumo
Introdução: A pancreatite da goteira (PG) constitui uma forma de pancreatite crónica segmentar, que afeta a área da goteira pancreatoduodenal, sendo frequentemente subdiagnosticada. O mecanismo fisiopatológico principal parece ser a obstrução ao fluxo da papila *minor* relacionada com o consumo de álcool. Esta patologia ocorre mais frequentemente em homens entre a 4ª e 5ª décadas de vida. A maioria dos doentes apresenta sintomas como náuseas e vómitos pós-prandiais, perda ponderal e dor

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abdominal. Apesar do desenvolvimento atual dos métodos radiológicos e endoscópicos, a distinção entre PG e neoplasia pancreática constitui um desafio diagnóstico e a avaliação histológica pode ser necessária. Se for possível obter o diagnóstico sem intervenção cirúrgica, o tratamento pode ser conservador na ausência de complicações agudas e crónicas. ***Apresentação do caso:*** Apresentamos 2 casos clínicos que demonstram a abordagem diagnóstica e a gestão de decisões terapêuticas nesta entidade. ***Discussão/Conclusão:*** A PG é uma entidade clínica que oferece com diagnóstico e terapêutica desafiantes. Apesar da importância crucial dos exames imagiológicos no diagnóstico e seguimento, a incapacidade de excluir um processo maligno torna necessária a intervenção cirúrgica numa parte significativa dos casos.

Introduction

Groove pancreatitis (GP), also described as paraduodenal pancreatitis, is a rare form of segmental chronic pancreatitis characterized by fibrotic scarring of the pancreatoduodenal (PD) groove, an anatomical area bound by the pancreatic head, duodenum, and common biliary duct (CBD) [1]. There are two forms of GP: pure GP occurring solely within the pancreaticoduodenal groove and segmental GP also affecting the pancreatic head [2, 3]. Its pathophysiology remains unclear, but it is likely to be multifactorial with a common pathway being the obstruction of the minor papilla [4]. Imagiological findings include fibrotic changes of the pancreatic groove and the presence of duodenal wall cysts as well as the thickening of the duodenal wall, pancreatic head enlargement, CBD, and Wirsung duct stricture. Given the clinical and radiological resemblance to pancreatic cancer, the diagnosis is challenging and frequently requires surgical intervention [5, 6]. We report two cases of GP as well as a review of the literature. Both our patients were male with a history of alcohol consumption and had a similar clinical and imaging presentation. However, they were managed differently with one of them requiring pancreatoduodenectomy.

Case Report 1

A 52-year-old male patient, with a history of hypertension and dyslipidemia, active smoking habits (35 pack units/year), and alcohol consumption over 100 g of alcohol daily, presented to the emergency department with right quadrant abdominal pain for over 5 months, with irradiation to the back associated with vomiting and weight loss (6% of the total body weight in 1 year). On examination, tender right hypochondrium palpation was noted. No signs of peritoneal reaction, ascites, or organomegaly were observed. Liver function tests, bilirubin, and amylase were within the normal ranges. He was referred to an outpatient consultation for follow-up.

The patient was reevaluated 5 months later, and because of his persistent abdominal pain and vomiting, an upper endoscopy was performed and revealed a bulging of the duodenal bulb with irregular mucosa. Duodenal biopsies were negative for neoplastic cells. Abdominal CT scan revealed diffuse parietal thickening of the second duodenal portion and diffuse densification of the surrounding tissues with preserved pancreatic structure and no Wirsung duct dilation (shown in Fig. 1). Serum carcinogen antigen 19.9 (CA 19.9) and IgG4 were normal. An abdominal magnetic resonance imaging (MRI) revealed heterogeneity in the pancreatic-duodenal recess, thickening of the second duodenal portion with a central cystic image (8 mm), and maintained pancreatic morphology and dimension (shown in Fig. 1). An endoscopic ultrasonography (EUS) showed hyperechoic foci and parenchymal lobularity, aspects of chronic pancreatitis, without pancreatic head nodular lesions or Wirsung duct dilation. Despite alcohol withdrawal, tobacco reduction, and pain medication, intense abdominal pain persisted with a great impact on the patient's quality of life. An abdominal CT was performed 14 months after the initial presentation, showing the same findings as before. The case was discussed at an oncology multidisciplinary meeting, and after discussion with the patient, he was submitted to a cephalic pancreaticoduodenectomy with complete remission of the abdominal pain. The pathology result was compatible with GP (shown in Fig. 2). At 5 years of follow-up, the patient is asymptomatic.

Case Report 2

A 63-year-old male with active smoking habits (60 pack units/year) and alcohol consumption (25 g alcohol per day) presented with a 1-month history of unintentional weight loss (31% of previous weight, BMI 15 kg/m^2) associated with postprandial vomiting and abdominal pain for 2 weeks. On examination, epigastrium and right hypochondrium tenderness on palpation were noted. He had no jaundice, palpable masses, or lymph nodes. Liver function tests, pancreatic enzymes, IgG4, and CA 19.9 were normal. Abdominal CT scan (shown in Fig. 3a) showed increased volume of the pancreatic head with small calcifications and a CBD of 8-mm upstream from the intrapancreatic portion. Upper endoscopy revealed extrinsic duodenal bulb compression (shown in Fig. 3b). EUS showed a heterogeneous pancreatic head parenchyma with hyperechoic foci with shadowing, but without Wirsung duct dilatation and duodenal wall thickening (25 mm). There was no evidence of nodular lesions (shown in Fig. 3c). The patient was maintained with nasojejunal feeding during 15 days with weight gain and pain relief.

The case was discussed at a multidisciplinary meeting, and it was decided to maintain the patient under active surveillance with conservative treatment. The patient stopped alcohol and tobacco consumption. Three months after discharge, he gained 18 kg and had no abdominal pain or vomiting. The MRI showed thickening and cystic changes in the duodenal wall and delayed enhancement in the PD groove, with a nondilated CBD. At 5 years of follow-up, he was asymptomatic and had normal laboratory tests.

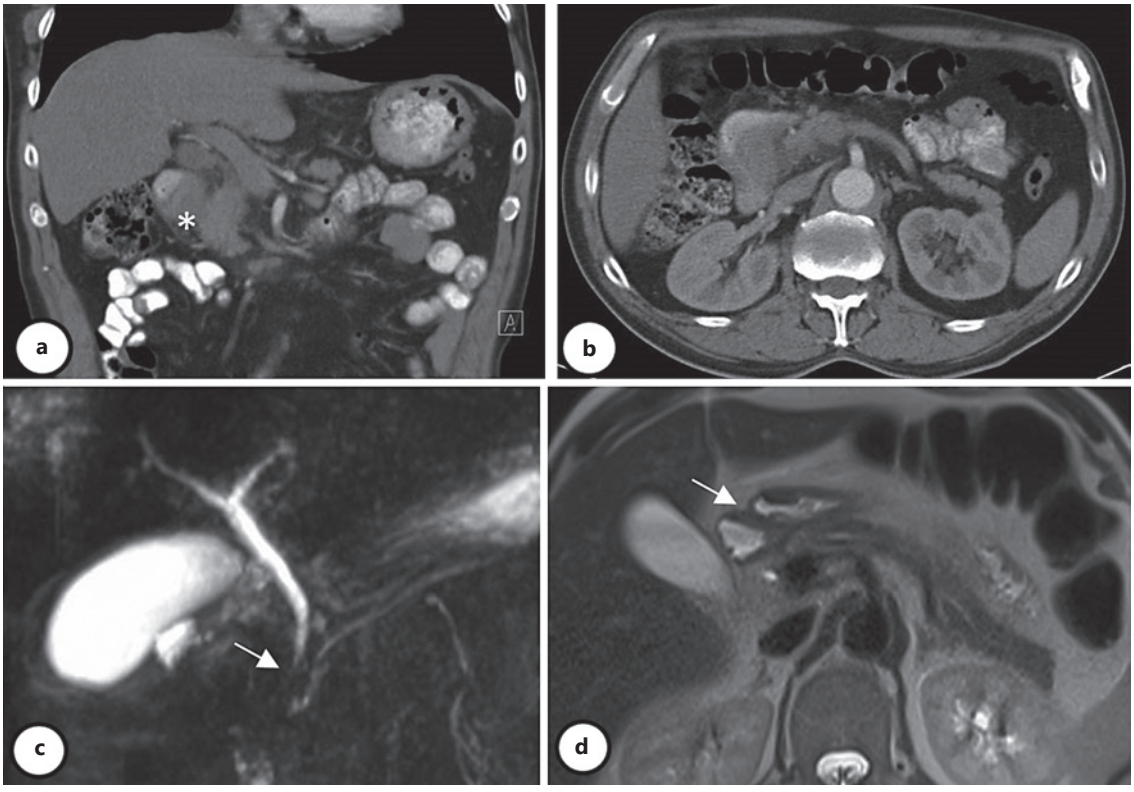

Fig. 1. Abdominal CT of case 1 showing a crescentic mass-like structure in the groove region (*) which causes duodenal stenosis (**a**) with a normal pancreas structure without Wirsung duct dilation (**b**) MRCP showing a smooth tapering of CBD (arrow) (**c**) and in a T2-weighted image (**d**) a cyst in the groove region. CT, computed tomography; MRCP, magnetic resonance cholangiopancreatography; CBD, common biliary duct.

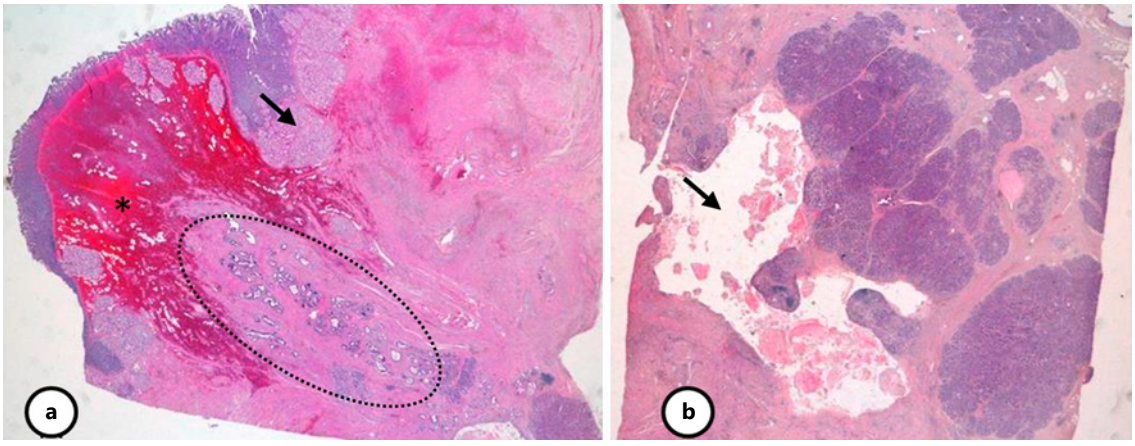

Fig. 2. Microscopic findings in case 1 showing (**a**). inflammatory changes with the presence of fibrosis and lymphoid infiltrate (circle), Brunner gland hyperplasia (arrow), and recent hemorrhage (*); (**b**) cystic spaces (arrow) and pancreatic acini with fibrosis.

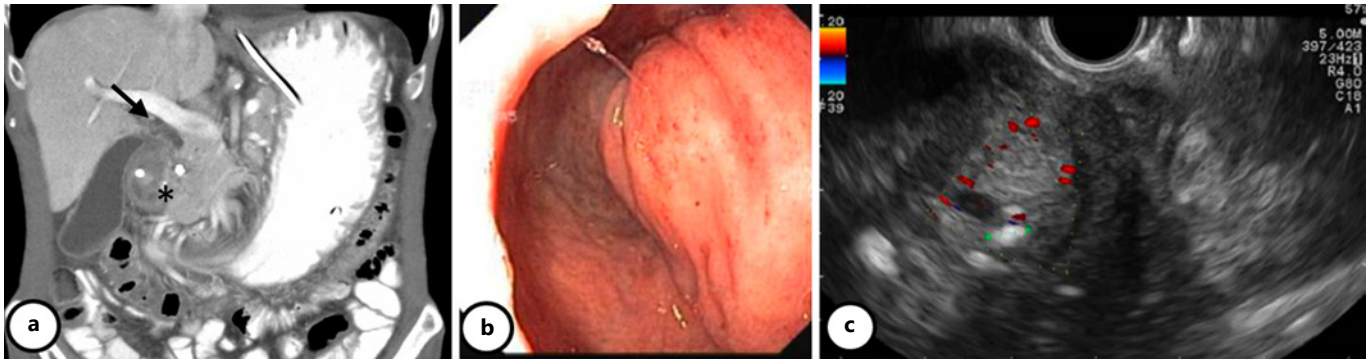

Fig. 3. a Abdominal computed tomography of case 2 showing enlargement of pancreas head with foci of cystic degeneration (*) and CBD dilation (8 mm) (arrow). **b** Upper endoscopy of case 2 showing duodenal stenosis (bulb represented) by extrinsic compression. **c** Endoscopic ultrasound of case 2 showing hypoechoic band-like thickening of the pancreaticoduodenal groove, as well as thickening of the adjacent duodenum and a hypoechoic heterogeneous pancreatic head. CDB, common biliary duct.

Discussion/Conclusion

GP – Pathophysiology

GP is a type of segmental chronic pancreatitis, first described in 1973 by Becker [2] with the German word Rinnenpankreatitis and translated into GP in 1982 by Stolte et al. [7]. The true incidence and its underlying etiology remain uncertain, but the central mechanism seems to be an anatomical or functional outflow obstruction of the minor papilla [1, 8–10]. The vast majority of patients have significant alcohol consumption, indicating that alcohol plays a major role in promoting pancreatic juice viscosity and exacerbating the inflammatory process [11]. Various factors contribute to outflow obstruction: pancreatic heterotopia in the duodenal wall with localized inflammation and duct dilation; Brunner gland hyperplasia, which can occur from chronic alcohol stimulation or from increased levels of cholecystokinin; or gastrin and pancreas *divisum* with subsequent increased pressure in the minor papilla [9, 12, 13].

Clinical Symptoms

GP manifests most commonly in men in their fourth to fifth decade of life with a history of chronic alcoholism, as occurred in our 2 cases [9]. The typical clinical symptoms are severe upper abdominal pain, nausea, recurrent postprandial vomiting, and weight loss [14]. Rarely, some patients can present with duodenal stenosis. Jaundice is rare and is often more suggestive of an underlying malignancy [10].

Diagnosis

The differential diagnosis includes pancreatic adenocarcinoma, periampullary cancers, pancreatic groove neuroendocrine tumor, autoimmune pancreatitis, and acute pancreatitis [3, 5]. Although in some patients, there are significant clinical and imaging features of GP, there is often overlap with other infiltrative processes involving the pancreatic groove, namely pancreatic cancer. Table 1 summarizes the clinical, laboratorial, and imagiological differences between GP and cancer.

Laboratory values and biochemical markers are often nonspecific. While liver function tests and bilirubin are generally within the normal range, alkaline phosphatase levels can be elevated even in the absence of biliary obstruction. Amylase and lipase may be slightly increased [4, 11]. Tumor marker's carcinoembryonic antigen and CA19.9 levels are generally normal. However, obstructive jaundice, if present, may cause an elevated level of CA 19.9, not related to an underlying malignancy [15]. IgG4 levels should be measured since autoimmune pancreatitis may mimic GP [16].

Imaging Features

Contrast-enhanced CT and MRI are the primary imaging modalities used when GP is suspected. The classic imaging features on CT scan consist of an ill-defined crescentic soft tissue mass seen in the PD groove with the pure form of GP. In its segmental form, there is a mass-like enlargement of the whole pancreatic head that can be indistinguishable from pancreatic cancer. The duodenal wall is involved in 92% of patients with luminal narrowing due to wall thickening. Smalls cysts are seen within

Table 1. Clinical, laboratory, and imaging features for distinguishing GP and pancreatic cancer arising in the groove region

	GP	Pancreatic cancer
Age	Younger patients (fourth–fifth decade)	Older patients
Ethanol abuse	Frequent	Uncertain association
Jaundice	Late event	Can occur early
Serum CA 19-9	Usually normal	Usually elevated
CT scan	Plate-like hypodense crescentic lesion and cysts in the PD groove	Round irregular pancreatic head mass
MRI	Sheet-like mass in the groove, hypointense on T1-weighted images, variable T2-weighted intensity. Cysts in the groove region	Round irregular pancreatic head mass
EUS	Duodenal wall thickening and luminal stenosis, long and smooth common bile duct stenosis, hypoechoic area and cysts in the groove region	Irregular and abrupt common bile duct stenosis, vascular encasement

CT, computed tomography; MRCP, magnetic resonance cholangiopancreatography; EUS, endoscopic ultrasound.

the duodenal wall or in the PD groove itself, in 81% and 75% of cases, respectively [17]. This common finding in GP contrasts with its rarity in pancreatic adenocarcinoma. CBD may be narrowed with a smooth, tapered, and regular stenosis [4, 11].The peripancreatic vessels are typically maintained, in contrast with the typical encasement in cases of adenocarcinoma. MRI shows a "sheet"-like mass of tissue, which is hypointense on T1-weighted images and variable in T2-weighted images, according to the time of disease onset [5]. Duodenal wall thickening is also seen, and T2-hyperintense cysts can be found in both the duodenal wall and PD groove [4, 9]. A study conducted by Kalb et al. [18] indicates that during focal thickening (>3 mm), abnormal increased enhancement of the second portion of the duodenum and cystic changes in the region of the pancreatic accessory duct are all present, the diagnostic accuracy is 87.2%, and a diagnosis of cancer can be excluded with a negative predictive value of 92.9%. Upper endoscopy typically shows mucosa edema, erosion, polypoid appearance of the descending duodenal part, and luminal stenosis [9]. EUS is considered by some authors the preferred imaging method as it provides high-resolution images of the head of the pancreas and PD groove, and it allows to obtain tissue sampling [5]. In GP, EUS can detect a hypoechoic band-like thickening of the PD groove and adjacent duodenum with intramural cysts, smooth stenosis of CBD and, in the segmental forms, a heterogeneous hypoechoic pancreatic head mass. Nonetheless, EUS is not able to differentiate inflammation and malignant infiltration in several cases [4, 11, 12].

Histopathology

Pathologic analysis of the pancreatic resection specimen is the only definitive way of diagnosing GP. Gross examination shows active and chronic inflammation of the PD groove, adjacent duodenal wall, and pancreatic head, as well as extensive scarring. Cystic spaces (0.2–2 cm) can be identified within the duodenal muscularis propria and submucosa and/or PD groove [9, 19].

The histological aspect of GP is characterized by thickening of the duodenal wall, inflammation of the Brunner gland and smooth muscle hyperplasia. Sometimes, heterotopic pancreatic tissue in the duodenal wall is identified, but this finding is not universal. Spindled stromal cells are the most common finding after fine needle aspiration (FNA). However, endoscopy-guided FNA biopsy presents great variability depending on the area sampled and the presence of cytological features associated with reactive cellular atypia resulting from pancreatitis may mimic neoplasia [4]. To our knowledge, there are no studies comparing EUS-guided FNA versus FNB, specifically in GP. However, recently, Wong et al. [20] compared the diagnostic performance of EUS-guided tissue acquisition by EUS-guided FNA versus EUS-guided FNB for solid pancreatic mass, and they found that the diagnostic yield of solid pancreatic mass was higher in FNB than in FNA (94.6 vs. 89.6%).

Treatment

Currently, there are no treatment guidelines on GP. When an accurate diagnosis of GP is possible, patients may be treated conservatively. Conservative therapy includes analgesia, alcohol and tobacco cessation, and parenteral nutrition when enteral nutrition is contraindicated, the patient does not meet the daily energy needs for more than 10 days, or in case of gastric or intestinal outlet obstruction [12]. There are also some reports of cases using somatostatin analogs, but the results appear to be temporary [21]. Endoscopic treatment may involve pseu-

docyst drainage, pancreatobiliary stent placement, or duodenal dilation. When cancer cannot be safely excluded, surgery is advised. Surgical treatment encompasses pancreatoduodenectomy, the most common procedure, and digestive or biliary bypass operations.

A recent systematic review evaluated clinical outcomes in GP after treatment [14]. The treatment was conservative in 29%, and half of the patients had complete symptom relief. Endoscopic and surgical treatment occurred in 12% and 59% of cases, respectively. Although surgery resulted in complete symptom relief in most of the patients (79%), it is associated with a high rate of complications (20%) [14]. Furthermore, in a large case series from Arvanitakis M et al., medical treatment associated with endoscopic approach (pancreatic ductal drainage, stricture dilation, and cyst drainage) allowed completed clinical success in 80% of the cases [22]. Thus, a stepwise approach, starting with conservative treatment, is recommended, unless malignancy cannot be ruled out.

In conclusion, we present the 2 cases of patients with classic risk factors, including alcohol and tobacco abuse, and symptoms of GP. Furthermore, they have similar clinical, laboratory, and imaging findings, namely, normal CA 19.9, parietal thickening of the descending part of the duodenum, and cystic changes without pancreatic duct dilation or vascular encasement. Thus, chronic abdominal pain and vomiting in a patient with history of alcohol abuse and no significant laboratory findings can point out to a chronic pancreatitis like GP. The treatment should be individualized, but a surgical approach may be necessary, as indicated in the first case.

Statement of Ethics

The authors have no conflicts of ethics. A written informed consent was obtained from participants for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Catarina Nascimento – acquisition and interpretation of clinical data for the case report, drafting the case report, and corresponding author. Carolina Palmela and António Soares – conception and design of the case, data collection, critically revising the case report, and final approval of the version to be published. Maria Lobo Antunes and Luísa Glória – critically revising the case report and final approval of the version to be published. Catarina Fidalgo – conception and design of the case, critically revising the case report, and final approval of the version to be published.

Data Availability Statement

All data generated or analyzed during this study are included in this article and/or its supplementary material files. Further inquiries can be directed to the corresponding author.

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