Clinical Case Report

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Intramural gastric pseudocyst

A case report and a comprehensive literature review

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Abstract

Rationale: Intramural pseudocyst, although first reported several decades ago, is a rare entity. Scientific knowledge regarding its clinical management is sparse.

Patient concerns: We present three cases to show the diverse clinical patterns of patients diagnosed with an intramural gastric pseudocyst.

Diagnosis: A final diagnosis should rest on proper evaluation by cross sectional imaging, including computer tomography and magnetic resonance imaging. Endoscopic ultrasound adds to the work-up.

Interventions: Previously, identified "lesions of the gastric wall" were not well recognized as an intramural pseudocyst, and treatments including resectional surgery were employed. Contemporary proper diagnostics should provide support to a less aggressive treatment approach.

Outcomes: While an indolent natural history without any clinical symptoms or discomfort could be expected in most cases, individual clinical evaluation should be applied.

Lessons: A heterogeneous information pattern from the limited number of cases in the literature makes it difficult to draw any firm conclusions. Attention to this rare condition should be increased to help clinicians arrive at a correct diagnosis and possibly prevent some patients from being over treated or from the use of unnecessary surgery.

Abbreviations: CT = computer tomography, ERCP = endoscopic retrograde choledocho-pancreatography, EUS= endoscopic ultrasound examination, MRCP = magnetic resonance choledocho-pancreatography, MT= magnetic tomography, RCC= renal cell carcinoma, US= ultrasound examination.

Keywords: cystic pancreas lesion, gastric lesion, intramural pseudocyst, pancreas, pancreatitis

1. Introduction

Intramural pseudocysts are uncommon; however, they have been reported in the literature for several decades.[1,2] The rarity of this condition and the paucity of reports in the scientific literature may contribute to the lack of awareness of this condition among clinicians, endoscopists, or radiologists caring for patients with various gastrointestinal symptoms or signs.

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Compliance with ethical standards

Informed consent to present pertinent medical information including imaging was obtained from the patients, and from the next of kin of the deceased patient. Publication is approved by the institution's research board.

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When cystic lesions are encountered on imaging, true pancreatic cystic lesions,[3] including the rare non-neoplastic cysts of the pancreas,[4] should be distinguished from pseudocysts to enable appropriate treatments for the proportion of patients in which interventions are necessary and beneficial.[5,6] However, the differential diagnosis of incidentally discovered cystic lesions on imaging of the upper abdomen are many and varied, including the rare occurrence of an extra-gastrointestinal stromal tumor presenting as an omental cyst,[7] a mature cystic teratoma of the pancreas,[8] and a spontaneous duodenal intramural hematoma,[9] to mention a few of the atypical diagnoses.

Pancreatic pseudocysts belong to a group of nonepithelial cystic pancreatic lesions because the epithelial lining is lacking.[10] Thus, pseudocysts, which are most commonly located in the peripancreatic region, are regarded as collections of pancreatic secretions enclosed in a fibrous tissue layer and mostly follow an episode of acute or recurrent pancreatitis.[10]

After a critical review of the pertinent literature on this topic and from the knowledge gained from 3 cases diagnosed with an intramural gastric pseudocyst encountered in our own practice, we want to address aspects regarding the clinical management of this rare condition.

2. The cases

2.1. Case #1

A 56-year-old multimorbid male (diabetes mellitus, atrial fibrillation, and depression) with a history of alcohol abuse for several years was admitted with unspecific abdominal complaints. His blood chemistry was normal, except for elevated

serum lipase (i.e., 4× the upper limit). An upper endoscopy was performed, and the results suggested a 2-cm submucosal lesion or impression at the fundus, with an otherwise negative examination. A gastric biopsy reported normal findings. Imaging with computer tomography (CT) revealed 2 cystic lesions related to the pancreas and slight atrophy of the gland with significant calcifications, which were consistent with sequelae evident of a previous episode of pancreatitis. Perigastric venous vessels in the upper abdomen were considered slightly dilated. Further imaging with magnetic tomography (MT) confirmed an atrophic pancreatic tail distal to a focal benign cystic lesion (diameter 37 mm) located in the corpus and another cystic lesion adjacent to the pancreatic tail (diameter 30 mm). The latter lesion involved the gastric wall and was consistent with an intramural gastric pseudocyst (Fig. 1). Using endoscopic ultrasound examination (EUS), a cystic intramural lesion (42 × 27 mm) of the cauda of the pancreas was identified (Fig. 1) as well as another cystic lesion (39 × 30 mm) that was located in the pancreatic corpus. In general, the pancreatic gland was regarded as atrophic with several tiny calcifications. A thin-walled gallbladder contained several stones, and the left adrenal gland was slightly enlarged; however, a tumor was not observed.

The patient's complaints resolved without any specific treatments, and attention was focused on optimizing his diabetes surveillance and treatment by his general practitioner in a context of significant socio-psychiatric challenges. Six months after the diagnosis of the intramural pseudocyst, the patient died unexpectedly at his house from a suggested cardiac arrest.

2.2. Case # 2

This case involved a male, 69 years of age, with a history of alcohol abuse and hypertension, who was surgically treated for prostate cancer. Over several years, the patient experienced repeated episodes of abdominal complaints and was diagnosed with relapsing pancreatitis based on biochemistry and imaging. The patient was previously treated endoscopically for duodenal hemorrhage. Upon recent admission to the hospital, the patient experienced pains and signs of gastrointestinal bleeding. The work-up, which included cross-sectional imaging (i.e., CT and US) and endoscopy revealed a bleeding area in the stomach, a pseudocyst with perforation into the lumen of the stomach, and a hematoma within the perforated pseudocyst (Fig. 2). Further investigations with repeated upper endoscopy and CT angiography did not show any ongoing bleeding. Supplementary magnetic resonance imaging (MRI) and a magnetic resonance choledochopancreatography (MRCP) of the pancreas showed patterns consistent with chronic pancreatitis, including parenchymal calcifications and dilatation and strictures of the pancreatic duct, and pseudocysts. Due to an upstream dilatation of the pancreatic duct, an endoscopic pancreatic stent was placed using ERCP, and the follow-up was uneventful.

2.3. Case # 3

This case involved a male, 73 years of age, who was treated successfully for an abdominal aortic aneurysm 2 years earlier; however, he developed significant postoperative renal insufficiency. Currently, his condition has been managed without the need for dialysis. Soon thereafter, a small (<20 mm in diameter) renal tumor was identified via cross-sectional imaging, and a clear Fuhrman grade 2[11] renal cell carcinoma (RCC) was confirmed on biopsy. Due to significant comorbidity, including his serious renal failure and the small-sized RCC with a low Fuhrman grade, a "wait-and-see" follow-up schedule was suggested to observe the tumor growth and the patient's general health condition. A subsequent re-evaluation of the previous cross-sectional imaging examinations over time revealed an incidentally discovered intramural gastric pseudocyst in this patient (Fig. 3). Careful re-appraisal of his

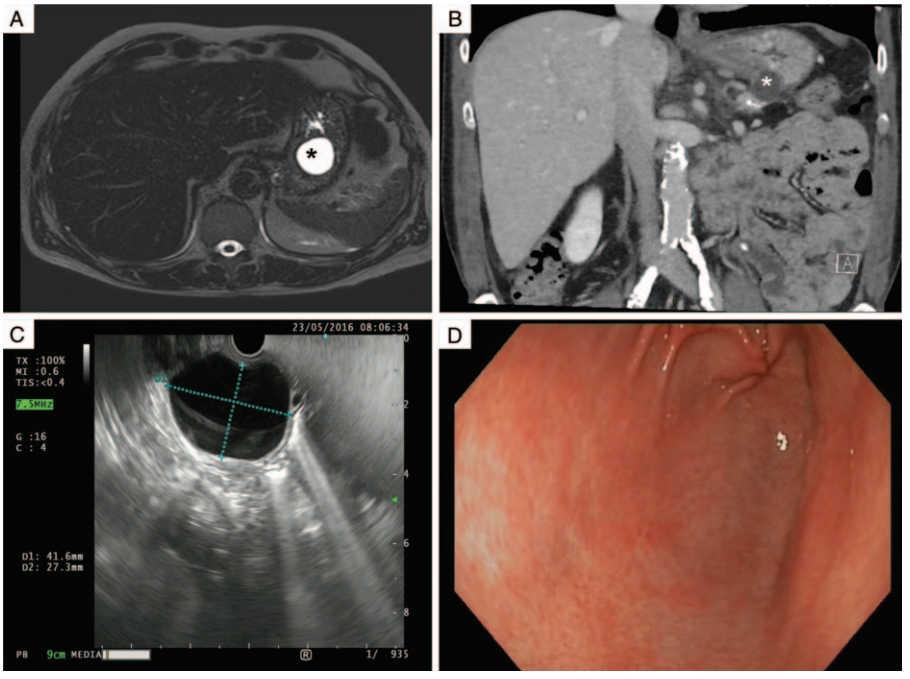

Figure 1. Patient # 1—Intramural gastric pseudocyst, (A) MRI with axial T2 shows a high signal in a well-circumscribed pseudocyst. (B) Coronal CT of the abdomen with intravenous contrast shows the intramural pseudocyst embedded into the stomach wall. (C) EUS shows the clear fluid content of the pseudocyst. The cyst appears to be located in the submucosal layer of the gastric wall. (D) Gastroscopy shows normal smooth mucosa found over the pseudocyst area. The pseudocyst is indicated with an*. CT = computer tomography, EUS = endoscopic ultrasound examination, MRI = magnetic resonance imaging.

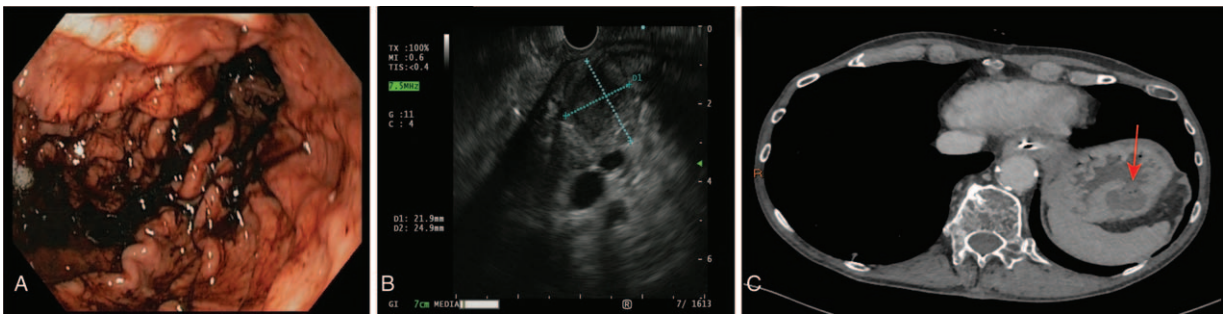

Figure 2. Patient # 2—Bleeding from an intramural pseudocyst with perforation into the stomach, (A) Gastroscopy shows bleeding and ulceration. (B) EUS shows a hematoma inside the perforated pseudocyst. (C) CT with intravenous contrast shows the perforation site (red arrow) of the pseudocyst comprising a heterogeneous hematoma. CT = computer tomography, EUS = endoscopic ultrasound examination.

clinical history confirmed episodes of abdominal pain, long-term alcohol abuse, and tiny pancreatic calcifications and pseudocysts shown on CT, which are all consistent with previous pancreatitis episodes. As shown in Figure 3, there is a slow growth of the intramural gastric pseudocyst over time. In this multimorbid patient without any pseudocyst-related symptoms, a watchful-waiting policy has been chosen along with the RCC surveillance conducted by the urologist.

3. Literature search

We used the terms " intramural cyst," "pseudocyst," "pancreatic cystic lesion," "gastric cyst," and "pancreatitis" in different combinations in an electronic PubMed search. We restricted the literature to the English language. Abstracts of identified references were reviewed to select articles that contained more than single case reports.

4. Results

Publications reporting more than single cases were scarce. A recent series from India comprising 9 patients (8 males), aged 24 to 54 years, and diagnosed between 2006 and 2013 was an exception.[12] These patients underwent a one-time endoscopic ultrasound (EUS)- guided aspiration of the pseudocyst, transmural drainage, transpapillary drainage, or surgery according to

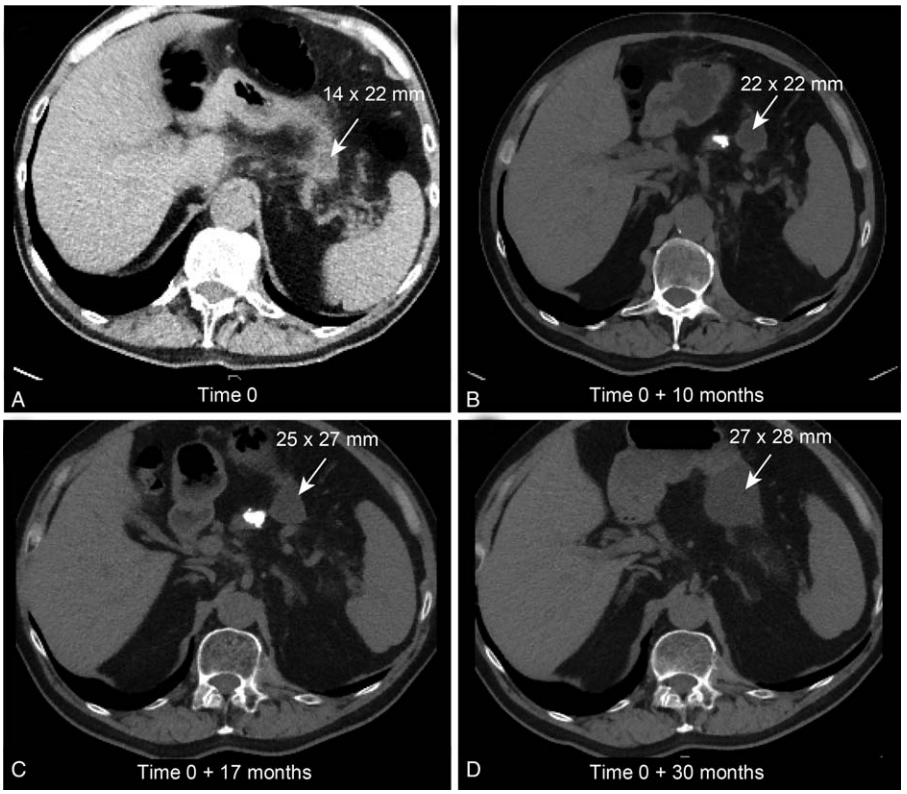

Figure 3. Patient # 3—An incidentally diagnosed intramural gastric pseudocyst, The identification of the pseudocyst was determined in retrospect by reevaluation of the CT imaging during follow-up of a nonsymptomatic patient after previous curative treatment for renal cell carcinoma. (A) The first available CT examination; white arrow indicates the intramural pseudocyst; (B) 10 months later; only minor cystic growth; (C) slight expansion after another 7 months; (D) the most recent CT performed 30 months after the first CT (see [A]) shows a continuous and slow growth pattern. The patient remains asymptomatic and progression-free of his right RCC. CT = computer tomography, RCC = renal cell carcinoma.

individual symptoms and lack of improvement after conservative management. Half of the patients had chronic pancreatitis, and the other half of the patients experienced acute pancreatitis. All patients complained of abdominal pain at presentation. The pseudocysts were located in the wall of the second part of the duodenum in 5 patients, within the gastric wall in 3 patients, and in the lower esophageal wall in 1 patient. The size of the pseudocysts ranged from 8 mm to 8 cm. In patients with duodenal intramural pseudocysts, symptoms suggestive of gastric outlet obstruction or jaundice were observed in addition to pain. The majority of the patients could be successfully treated endoscopically with no significant complications. In a previous report, Oka and coworkers[13] presented accumulated data from 9 case reports of gastric intramural pseudocysts related to pancreatitis published between 1966 and 2007. All patients were men, with a median age of 41 (range 20–68) years. The etiology was alcohol in at least 8 of the 9 patients. While surgery was employed in most cases, internal drainage and spontaneous internal drainage occurred in 2 patients.

Our 3 male patients, aged 52–72 years, were diagnosed within the last two years and had alcohol as a common etiologic factor. One patient had been treated for acute gastrointestinal bleeding, which was suggested to be related to pseudocyst perforation to the stomach lumen, and eventually received a pancreatic stent for ductal stricture. The remaining two patients were followed-up conservatively.

5. Discussion

A suspicious finding of a "lesion" on upper endoscopy or on cross-sectional imaging may be more or less obvious;[14] however, the recognition and interpretation of this finding as an intramural gastric or duodenal pseudocyst may be a challenge.[6,15,16] This rare condition, although first described approximately 50 years ago,[2] does not seem to be easily recognized or interpreted correctly, which may cause diagnostic delay or inappropriate management.[4,5,17]

Most patients with intramural pseudocysts are men, and a history of pancreatitis is common,[12,13] as illustrated in our patients. Abdominal complaints are frequently reported, and symptoms and signs of gastric outlet syndrome or jaundice are also encountered.[18,19]

Using modern imaging, including CT, MRI, and EUS, the diagnostic challenge to separate a "suggestive lesion" from a true solid tumor should be achieved.[3,4,6,16] While a history of pancreatitis is frequently encountered in patients with an intramural gastric pseudocyst, other etiological factors including the rare occurrence of heterotopic pancreatic tissue should also be considered.[18,20] The accurate localization of a cystic intramural lesion combined with a history of pancreatitis may help make a correct diagnosis. Nevertheless, as also reported by us more than 2 decades ago, the close connection between an "ordinary" pancreatic pseudocyst and the stomach wall may cause symptoms and severe complications (i.e. gastric haemorrhage).[21] Thus, one can of course speculate if our present case #2 had a true intramural or "an ordinary" pseudocyst. By careful examination of all available information, including imaging, we think this is an intramural location.

Recent changes in the terminology of fluid collections in acute and chronic pancreatitis have prompted novel suggestions as to the evolution and outcomes of pseudocysts. When pseudocysts or acute peripancreatic fluid collection occur, as recently defined by the revised classification of acute pancreatitis,[10] more than one-half of these will resolve without any intervention within 6 months.[22] Honselmann and coworkers[5] recently reported that a high preoperative diagnostic accuracy (95%) was achieved when a benign lesion was suspected, whereas only 42% of the lesions were predicted correctly. Thus, in asymptomatic patients with suggested benign focal cystic lesions, a wait-and-see approach would be preferable when a diagnosis of an intramural gastric pseudocyst is considered. When symptoms (i.e., abdominal pain, nausea, and vomiting) or a sign of biliary obstruction occur related to a particular finding on cross-sectional imaging, therapeutic approaches including EUS-guided drainage should be employed according to the updated guidelines.[16,17] We did not recognize any specific indications which should prompt an EUS-guided drainage for diagnostic or therapeutic reasons in our 3 patients. However, others may think a more proactive approach would be appropriate in this clinical decision. The diagnostic work-up, which is mainly based on cross-sectional imaging (i.e., CT and MR), could include EUS when clinically indicated to arrive at a diagnosis that can accurately distinguish between benign and malignant lesions. This recommendation would direct the clinical management toward the prevention of overtreatment or the use of unnecessary surgery.[5,17]

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