

Original Article

Intramural Pseudocysts of the Upper Gastrointestinal Tract

Surinder Singh Rana^{1*}, Deepak Kumar Bhasin¹, Chalapathi Rao¹, Rajesh Gupta²

Departments of ¹Gastroenterology and ²Surgery, Post Graduate Institute of Medical Education and Research, Chandigarh, India

Abstract

Background: Intramural pseudocysts or pseudocysts occurring in the gastrointestinal tract (GIT) wall are rare and there is a paucity of data on their clinical features and management.

Patients and Methods: We retrospectively evaluated patients with intramural pseudocysts seen at our institution over the past 6 years. Intramural location was confirmed either on surgery or endoscopic ultrasound (EUS). Depending upon the symptoms, their control with the conservative medical therapy and procedural consent, the patients underwent EUS guided single time aspiration of the pseudocyst or transmural drainage or transpapillary drainage or surgery.

Results: A total of 9 patients with intra mural pseudocysts (male: $n = 8$; mean age \pm SD: 39.3 ± 8.0 years; age range: 24-54 years; five patients having chronic and four patients having acute pancreatitis) were studied. The pseudocysts were located in the wall of the second part of the duodenum in five patients, in the gastric wall in three patients and in the lower esophageal wall in one patient. The size of the pseudocysts ranged from 8 mm to 8 cm and 3/9 (33%) patients had associated extra mural pancreatic pseudocysts. All patients had abdominal pain on presentation. Along with pain the patients with duodenal intramural pseudocysts also had symptoms suggestive of gastric outlet obstruction (3) or jaundice (1). The patient with esophageal intramural pseudocyst had dysphagia along with abdominal pain. Majority of these patients could be successfully treated endoscopically with no significant complications.

Conclusions: Intramural pseudocysts of the upper GIT are very rare and EUS is a useful investigational modality for diagnosing and treating them.

Keywords: pseudocysts; pancreas divisum; endoscopic ultrasound; papillotomy

Rana SS, Bhasin DK, Rao C, Gupta R. Intramural Pseudocysts of the Upper Gastrointestinal Tract. *Endosc Ultrasound* 2013; 2(4): 194-198.

INTRODUCTION

Pancreatic pseudocysts are a collection of pancreatic secretions enclosed in fibrous tissue layer without a lining of epithelium and are usually located in the peripancreatic region.¹ They may also rarely occur at unexpected and atypical locations such as the spleen, liver, mediastinum, pelvis and kidney depending upon the path taken by the activated pancreatic enzymes.^{1,2}

Intramural pseudocysts or pseudocysts occurring in the gastrointestinal tract (GIT) wall are very rare. They have been reported in the stomach; duodenum and colon as occasional case reports.³⁻¹³ The exact mechanism of the formation of the pseudocysts in the GIT wall are not

known. The possible mechanisms are suggested for their formation includes rupture of a pancreatic pseudocyst into the GIT wall, presence of a fistula between the pancreas and the alimentary tract and inflammation of heterotopic pancreatic tissue within the GIT wall.³⁻¹³ Due to their rarity there are no guidelines on their management and most of the described cases in the literature have been either treated surgically or have spontaneously decompressed by rupturing into the GIT lumen.³⁻¹³

In this study, we describe the clinical and radiological characteristics of intramural pseudocysts in nine patients as well as our experience with endoscopic drainage and clinical outcome in these patients.

PATIENTS AND METHODS

We performed a retrospective analysis of patients with intramural pseudocysts seen at our institution over the past 6 years. Clinical records were reviewed to identify patient

*To whom correspondence should be addressed.

E-mail: drsurinderrana@yahoo.co.in

Received: 2013-04-03; Accepted: 2013-05-04

DOI: 10.4103/2303-9027.121240

symptoms and imaging findings. All patients were symptomatic and had intramural pseudocysts with a well-formed wall, as documented on contrast enhanced computed tomography (CECT) scan. The intramural location was confirmed either on surgery or endoscopic ultrasound (EUS). The EUS examination was performed with either a radial scanning echoendoscope (EG-3670 URK radial echoendoscope, Pentax Inc., Tokyo, Japan) or a linear scanning echoendoscope (EG-3870 UTK linear echoendoscope, Pentax Inc., Tokyo, Japan) at 7.5 MHz. Depending upon the symptoms, their control with the conservative medical therapy and procedural consent, the patients underwent EUS guided single time aspiration of the pseudocyst or transmural drainage or transpapillary drainage or surgery. All symptomatic patients with less than 3 cm size pseudocysts were initially treated by a single time EUS guided aspiration. Following this, if the symptoms persisted or the pseudocysts recurred, patients were treated endoscopically or by surgery. All patients provided procedural informed consent at the time of EUS examination or the endoscopic treatment.

For EUS guided aspiration or endoscopic drainage, intravenous ciprofloxacin was administered for antibiotic prophylaxis. EUS guided single time aspiration was performed using a 19 G (in the stomach) or 22 G (in the duodenum) needle. Endoscopic retrograde pancreatography was performed by standard technique using a TJF 145 or TJF 160 (Olympus Optical Co. Ltd., Tokyo, Japan) side-viewing duodenoscope under conscious sedation using intravenous midazolam. Hyoscine butyl bromide was used to inhibit duodenal peristalsis. Pancreatic duct (PD) disruption was defined by free extravasation of contrast outside the PD system as seen on fluoroscopy after retrograde contrast injection of the main PD or dorsal duct (in patients with pancreatic divisum). PD disruption was defined as complete when the main duct upstream to the disruption was not visualized on fluoroscopy and as partial when the main duct was visualized upstream from the site of disruption. After confirming the ductal disruption, a 5-F stent was placed across the papilla into the PD by advancing it over a 0.025- or 0.035-inch hydrophilic guide wire (Jagwire) (Microvasive Endoscopy,

Boston Scientific Corp., Natick, MA). An attempt was made to place the stent across the area of disruption.

Therapeutic success was defined as symptomatic improvement with radiological resolution of all pseudocysts on CECT scan and therapeutic failure was defined as persistence of pseudocyst at 8 weeks after endoscopic therapy or need for surgical or radiological intervention. Following resolution, the stent was removed and a repeat pancreatogram was obtained to document healing of ductal disruption.

RESULTS

A total of 9 patients with intra mural pseudocysts (male: $n = 8$; mean age \pm SD: 39.3 ± 8.0 years; age range: 24-54 years) were seen by us over past 6 years (Tab. 1). Five patients had chronic pancreatitis and four patients had pseudocysts as sequelae of acute pancreatitis. Majority of the patients (8/9; 89%) had alcoholic pancreatitis (chronic five and acute three) and one patient also had associated complete pancreas divisum. The only female patient developed intramural pseudocyst as a consequence of acute gall stone pancreatitis. The pseudocysts were located in the wall of the second part of the duodenum in five patients, in the gastric wall in three patients and in the lower esophageal wall in one patient. The size of the pseudocysts ranged from 8 mm to 8 cm and 3/9 (33%) patients had associated extra mural pancreatic pseudocysts.

All patients had abdominal pain on presentation. Along with pain the patients with duodenal intramural pseudocysts also had symptoms suggestive of gastric outlet obstruction (3) or jaundice (1). The jaundice developed because of compression of the bile duct by the duodenal pseudocyst. Patient with esophageal intramural pseudocyst had dysphagia along with abdominal pain.

All the gastric and duodenal pseudocysts were well-demonstrated on CECT (Figs. 1 and 2). The patient with esophageal pseudocyst had thickening of the lower esophageal wall demonstrated on CECT, but no definite pseudocyst could be visualized. However, EUS demonstrated small intramural pseudocyst with wall thickening and loss of wall stratification

Table 1. The profile of nine patients with intra mural pseudocysts

Age/sex	Etiology	Acute/chronic	Location	Predominant symptom	Size (cm)	Extra mural pseudocyst	Management
36/M	Alcohol	Chronic	Duodenum	GOO	1.2	No	Aspiration
42/F	Gallstones	Acute	Duodenum	EHBO	1.4	No	Aspiration+biliary stent
38/M	Alcohol	Chronic	Duodenum	GOO	8	No	Surgery
42/M	Alcohol	Chronic	Duodenum	GOO	2	No	Aspiration
54/M	Alcohol	Acute	Stomach	Pain	6	No	Conservative
24/M	Alcohol	Chronic	Stomach	Pain	2	No	Aspiration+minor papillotomy
36/M	Alcohol	Acute	Stomach	Pain	4	Yes	Conservative
38/M	Alcohol	Acute	Duodenum	Pain	1.5	Yes	Conservative
44/M	Alcohol	Chronic	Esophagus	Pain with mild dysphagia	0.8	Yes	Transpapillary drainage

M: Male; F: Female; GOO: Gastric outlet obstruction; EHBO: Extra hepatic biliary obstruction

(Fig. 3). EUS was also done in other seven patients and it could clearly demonstrate intra mural pseudocyst in all these seven patients (Figs. 1, 2 and 4). One of these patients had significant necrotic debris within the pseudocyst. None of these patients received parenteral nutrition or octreotide or somatostatin.

One patient with a large duodenal pseudocyst causing gastric outlet obstruction preferred surgery and was treated surgically. All the remaining patients with duodenal pseudocysts and gastric outlet obstruction ($n = 2$) underwent EUS guided aspiration of the cyst with a 22 G needle and it revealed hemorrhagic fluid with markedly elevated amylase and lipase and normal carcino embryonic antigen levels. The cyst was completely emptied and a nasojejunal tube was placed for enteral feeding. The oral feeding was gradually reintroduced and once patient tolerated oral feeds well the

nasojejunal tube was removed. The patient with obstructive jaundice underwent single time EUS guided aspiration of the pseudocyst along with an insertion of a biliary stent that was removed after 4 weeks. One patient with small duodenal pseudocyst and pain only was successfully treated by medical management of oral enzymes, anti-oxidants and non-steroidal anti-inflammatory drugs.

One of patients with gastric pseudocyst was treated with a combination of a single time EUS guided aspiration along with minor papillotomy. The remaining two patients with gastric pseudocyst were successfully managed with medical therapy alone. The patient with esophageal pseudocyst was successfully treated with endoscopic transpapillary drainage

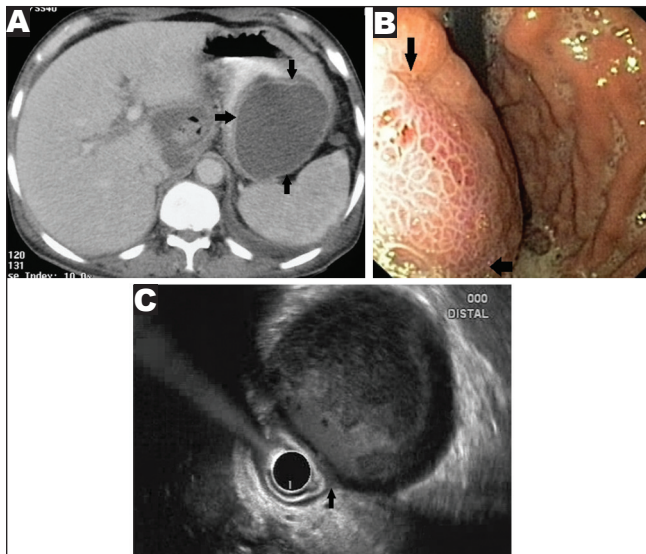


Figure 1. (A) Contrast enhanced computed tomography: Large gastric intramural pseudocyst (arrows); (B) endoscopic image: Submucosal bulge in stomach; (C) endoscopic ultrasound: Intra mural pseudocyst adherent to the muscularis propria (arrow) of the gastric wall

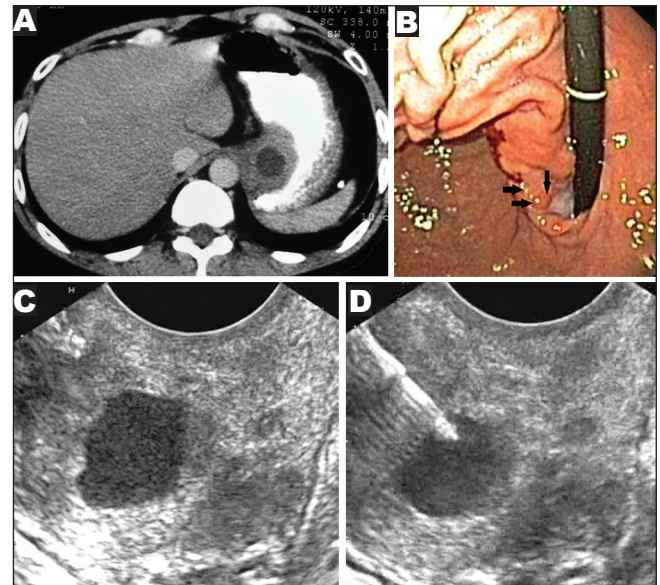


Figure 2. (A) Contrast enhanced computed tomography: Gastric intra mural pseudocyst; (B) endoscopic image: Nodularity just below gastro esophageal junction (arrows); (C) endoscopic ultrasound (EUS) intramural pseudocyst with gastric wall thickening and loss of wall stratification; (D) EUS guided aspiration of intramural pseudocyst

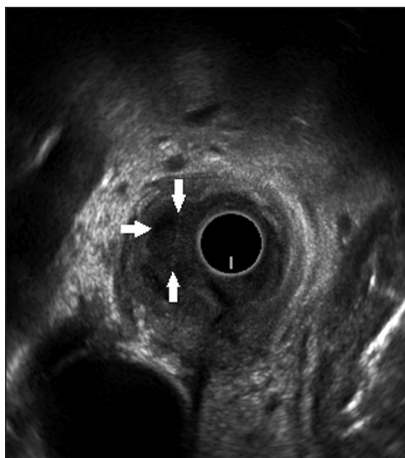


Figure 3. Esophageal intra mural pseudocyst with wall thickening (arrows)

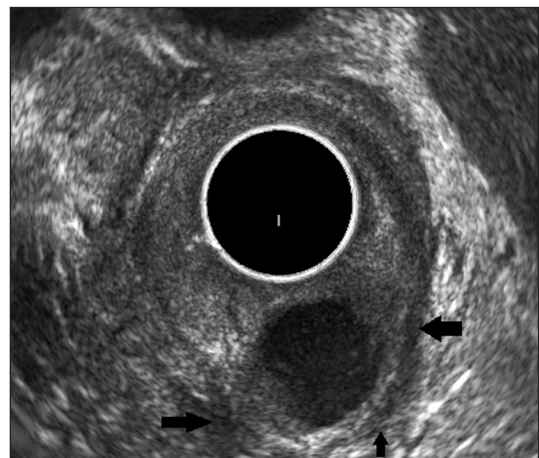


Figure 4. Duodenal intramural pseudocyst. Muscularis propria seen around the cyst (arrows)

using a 5 Fr stent. This patient had dilated main PD along with partial disruption in the body of the pancreas.

No significant complication of the procedure was noted in any of the patients. In a follow-up period of 2 months to 6 years, there has been no recurrence of symptoms in these successfully treated patients.

DISCUSSION

Intramural pseudocysts of the GIT are very rare and have been reported in the stomach, duodenum and colon.³⁻¹³ The rarity of intramural pseudocysts suggests that the GIT wall seems to be a relatively strong barrier to proteolytic activity of pancreatic enzymes. However, once the barrier is broken, the expansion of the intramural pseudocyst can lead to obstruction of the lumen and pain as was seen in the majority of our patients.³⁻¹³ With accumulation of pancreatic secretions these pseudocysts may extend within the wall or may rupture into the bowel lumen.

Intramural gastric pseudocysts are very rare and a literature review in 2003 revealed seven published cases of gastric intramural pseudocysts.¹⁴ The exact mechanism of formation of gastric pseudocyst is not known and the suggested possibilities include rupture of pseudocyst into the wall of the stomach, presence of pancreaticogastric fistula and pancreatitis occurring in heterotopic pancreatic tissue within the gastric wall.¹⁴ The endoscopic appearance of gastric intramural pseudocyst resembles any gastric submucosal lesion as was in our cases (Figs. 1 and 2). The CECT is an useful investigation for confirming the cystic lesion of the lesion as well detecting features of acute or chronic pancreatitis.^{10-12,14} However, EUS is the most useful investigation for evaluating patients with intramural pseudocysts. It can help in evaluating the contents of the cyst in detail, excluding significant necrotic debris and ascertain the relationship of the cyst with the various layers of the GIT wall.^{10,15} Aspiration of fluid high in amylase and lipase content is also a reliable modality for confirming the diagnosis of intra mural pseudocyst as was done in one of our case.⁹

The duodenal intramural pseudocysts have also been rarely reported and they usually occur posteriorly with the second part of the duodenum.^{3,5} This is because the posterior surface of the duodenum is in direct contact with the head of the pancreas with no barrier to prevent the digestive effects of pancreatic secretions.^{3,4} Depending on the depth of the penetration these duodenal pseudocysts may develop between the serosa and muscularis, or between muscularis and mucosa. The differential diagnosis for such duodenal lesions includes a duodenal duplication cyst and a choledochocoele. Careful history and investigations such as CECT and EUS can help in accurately diagnosing intramural pseudocyst.

In patients with duodenal intra mural pseudocysts the symptoms of gastric outlet obstruction dominate the clinical feature as was also in the majority of our patients.^{3,5} The

role of endoscopy in diagnosing intra mural pseudocyst is limited as its endoscopic appearance resembles any duodenal submucosal lesion. CECT is an useful investigation for diagnosis of duodenal intra mural pseudocysts and the CT characteristics of intramural involvement are an extension of the pseudocyst along the course of the duodenum and flattening of the pseudocyst wall at the border of the GIT lumen.⁴ However, like for gastric intra mural pseudocysts, EUS is the most useful investigation for evaluating patients with duodenal intramural pseudocysts.⁵

Due to their rarity, there is no consensus on the best approach for their management. Majority of published cases either have been diagnosed at laparotomy or treated by surgical decompression.³⁻¹⁵ However, as the intramural pseudocysts are immediately adjacent to the GIT lumen, they can be effectively treated by endoscopic aspiration/drainage as was done in the majority of our cases.

CONCLUSION

Based on the study it can be concluded that intramural pseudocysts of the upper GIT are very rare and EUS is the most useful investigational modality for diagnosing and treating them. Gastric intramural pseudocysts frequently present with abdominal pain and symptoms of gastric outlet obstruction dominate the clinical presentation of patients with duodenal intramural pseudocysts.

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