

Paravaterian diverticula presenting as acute cholangitis in two very old patients

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Citation

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Abstract

Paravaterian diverticula are extraluminal outpouchings of the duodenal lumen arising in a range of 2-3 cm from the ampulla of Vater. Their prevalence increases with age. Complications include pancreaticobiliary complications (primary calculosis of the biliary tract; cholangitis; acute pancreatitis) and non-pancreaticobiliary complications (bleeding; perforation; obstruction). The association between diverticula and the listed pathologies is not univocal and the pathophysiology that leads to the single pancreato-biliary complication is not well understood. We report two cases of very old patients presenting with epigastric pain, fever and a laboratory pattern of acute cholangitis related to the presence of paravaterian diverticula.

FIRST CASE

A 92-year-old man was admitted to the hospital because of an epigastric pain. Ten days before admission the patient developed constrictive pain, initially not well localized, but prevalent in epigastrium, continuous, of intensity 8 on a scale of 0 to 10, where 10 is the most severe, inconstantly radiating to the left shoulder, not accompanied with other symptoms.

His medical history included chronic obstructive pulmonary disease, coronary heart disease with a previous MI 10 years before, treated at that time with PTCA + stenting on LAD, a *Helicobacter pylori*-related gastritis eradicated 4 years before.

On examination, body temperature was 37,2 °C, there were skin and mucosal signs of dehydration, the abdomen was mildly dilatated for fat's accumulation, tender on deep palpation in epigastrium and right upper quadrant, peristalsis was present, no signs of peritonism were evident. There was mild hepatomegaly with the lower liver margin palpable at 4 cm from the ribs, and was smooth and slightly painful. The reminder of examination was normal.

The diagnostic hypothesis included acute coronary syndrome, acute gastritis, peptic ulcer disease, GERD, acute pancreatitis, cholecystitis, biliary colic and cholangitis.

During the first days troponin I and myoglobin were normal on repeated controls, 12 lead ECG was repeatedly normal, thus excluding a cardiac etiology of the pain.

On the second day after admission the patient developed jaundice, fever and recurrence of epigastric pain, continuous and without features of colic. Blood tests showed an increase of the indices of cholestasis (gGT 114 U/L, bilirubin tot. 147,2 umol/L conjugated bilirubin 104,2 umol/L), of indices of hepatic cytolysis (AST 290 U/L ALT 208 U/L) and of the indices of inflammation (Leucocytes 13,70 x10⁹/L, neutrophil 87% PCR 183 mg/L), amylase and lipase and were normal, thus excluding acute pancreatitis. A blood culture grew *Klebsiella Pneumoniae*.

An ultrasound of the upper abdomen showed liver of normal size, shape and structure, the biliary tree was mildly dilatated, the terminal part of the common bile duct was not visible, and the gallbladder was dilatated, with regular walls and echogenous material like biliary sand inside.

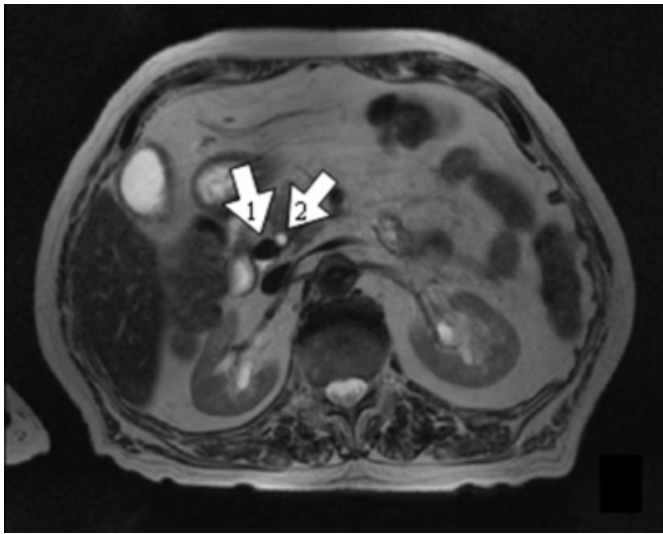
In consideration of the clinical picture (triad of Charcot), of blood and imaging tests, a diagnosis of acute cholangitis was made. Antibiotic therapy with ceftriaxone and intravenous fluids were started. In the following days a steady improvement of jaundice, fever and abdominal pain was observed, bioumoral controls showed a progressive decrease of cholestasis and inflammation indices.

A nuclear magnetic resonance of the upper abdomen with colangio-NMR showed the presence of a paravaterian diverticulum located cranially to the papillary outlet in the duodenal lumen, the biliary tract was regular without filling defects (calculi), gallbladder walls were thickened, the

pancreatic duct was regular [Picture 1].

Considering the age and the absence of signs of bleeding, perforation or obstruction open surgery was not indicated; We discussed whether to perform or not an ERCP which was actually excluded due to the positive clinical evolution with medical therapy, and the absence of clear evidence of long term benefits in literature. At 1 year follow-up the patient remains disease free and there has been non recurrence of pancreaticobiliary symptoms.

Figure 1



Picture 1: Abdominal Cholangio-NMR - T2 weighted images: arrow 1 highlights the duodenal diverticulum, in its lumen the contrast medium forms a little hydroaereal level; arrow 2 indicates the mildly dilated common bile duct.

SECOND CASE

A 92-year-old woman was admitted to the hospital because of a serious and progressive anaemia (Hb from 10 to 8,6 mg/dl). Three days before admission she developed intermittent fever with peak temperature up to 37,8 C°. At the Emergency Department temperature was of 39°C, blood test revealed AST 190 U/L, ALT 64 U/L, total bilirubin 21,8 umol/L and CRP 27,7 mg/L.

Her medical history included ischemic-hypertensive cardiopathy, cognitive impairment on cerebrovascular base leading to a hypokinetic syndrome, several hospitalizations for coprostasis and sub-occlusion, an episode of DVT 3 years before. She had been admitted to another hospital 3 months before because of the development of common bile duct calculosis. At that time an ERCP with endoscopical papillotomy and removal of multiple gallstones had been

performed.

On examination the patient was pyretic, sweaty, confused, blood pressure was 110/60 mmHg, heart rate 80, SatO₂ 90% while breathing ambient air. On cardiac auscultation a systolic murmur of 2/6 of intensity was found. The abdomen was tender in right hypochondrium, the peristalsis was slowed. The remainder of examination was normal.

A chest X-ray evidenced pleural effusion and density in the left lower lung field, such findings were however already present in a previous X-ray film 4 months before. An ECG showed sinus rhythm with FC 80/min, QRS axis + 30° and normal ventricular repolarization.

On the basis of the strong clinical suspicion of a new episode of choledocolitiasis, complicated with cholangitis, a liver ultrasound was performed: the biliary tree was dilated, several calculi were visible in the common bile duct, while there was no evidence of calculi in the gallbladder.

The patient was conservatively treated with antibiotics (intravenous ceftriaxone) and intravenous fluids. The persistence after four days of fever, abdominal pain, and abnormal liver function tests induced to perform a new ERCP. During the procedure new gallstones in the main bile duct were removed, the presence of a duodenal diverticulum located cranially to the papilla of Vater was noticed. On the basis of the antibiogram of blood culture (*Staphylococcus Epidermidis*) antibiotic therapy was changed introducing levofloxacin and trimethoprim-sulfamethoxazole.

The ERCP had no complications and the reminding of the hospital stay was uneventful, the patient became afebrile and asymptomatic of the abdominal pain, with prompt normalization of liver test, and recovered in ten days. At 1 year follow-up the patient had not suffered any new episode of cholangitis.

DISCUSSION

Periampullary diverticula (PAD), also called paravaterian diverticula are extraluminal outpouchings of the duodenal lumen arising in a range of 2-3 cm from the ampulla of Vater. The true prevalence of PAD in the general population has not been defined in an univocal way, the prevalence reported ranges from 0.16% to 27% [1], depending on the different clinical setting and the different imaging technique used. The prevalence of PAD increases with age [1]: the real reasons of the correlation between age and prevalence of PAD are not completely clear, it is however possible to

speculate that with the population ageing PAD may become more frequent than previously thought: we point out that both above described patients were 92 years old.

The complications that are potentially associated with PAD are listed in Table 1. The association between diverticula and the listed pathologies is not univocal and the pathophysiology that leads to the single pancreatobiliary complication is not well understood. The two main causes seem to be the motility dysfunctions of the sphincter of Oddi and the predisposition to the bacterial contamination of the biliary tract.

Figure 2

Table 1: Complications potentially associated with periampullary diverticula.

Pancreaticobiliary complications	Non-pancreaticobiliary complications
Primary calculosis of the biliary tract Cholangitis Acute pancreatitis	Bleeding Perforation Obstruction

Studies conducted with endoscopic manometric techniques suggest that the presence of PAD is associated with dysfunctions of the papillary motility (perhaps related to a prolonged mechanical compression of the sphincter), a diminution in sphincter tone and an increase in intrabiliary pressure [2-4].

PAD, like other blind pouches of the gastrointestinal tract, favour bacterial overgrowth, patients with PAD have positive bile culture more frequently, most common microorganisms are enterobacteriaceae like E.Coli, Klebsiella spp., Proteus spp., S. Fecalis. [5].

The association between PAD and the formation of pigmented gallstones in the biliary tract has been confirmed in several studies. As mentioned above the two mechanisms explaining this association are papillary dysfunction and bacterial contamination: the colonization of the biliary tract from glucuronidase-producing bacteria favours the deconjugation of the bile and its precipitation in insoluble stones of calcium-bilirubinate. The presence of PAD also predisposes to the recurrence of calculosis, colecistectomy does not prevent the recurrence [6-7].

The association between the presence of PAD and cholangitis is more debated: there is no doubt that cholangitis can occur secondarily to the calculosis of the biliary tract, as happened in the second case presented. On the other hand the possibility that paravaterian diverticula can cause cholangitis per-se (i.e. in the absence of

calculosis) is more controversial. Such an occurrence has been defined Lemmel’s syndrome. Recent studies [8-10] support this possibility: the hypothesized mechanisms are the alterations of the papillary motility, the bacterial contamination and the compression ab extrinseco of the main biliary tract.

The presence of PAD has also been implicated with the occurrence of acute pancreatitis: the bases of an eventual association are physiopatologically grounded, and are similar to those discussed for cholangitis; however, the evidence available from the literature is still limited and insufficient in to demonstrate a directed association in the absence of calculosis [11-12].

The therapeutic options for PAD include surgical diverticulectomy, endoscopic retrograde cholangiopancreatography (ERCP) with papillotomy and a conservative attitude.

Open-surgery of diverticula is currently less used and there is no proven long term benefit in patients with pancreaticobiliary symptoms (differently from patients with diverticulum perforation or bleeding) [1] and most reports are of single cases or very small selected series [13]; for the clinical characteristics of the cases described above, surgery has never been a true alternative for the management of those patients, and is not further discussed in this paper.

The presence of diverticula arising close to the ampulla can make the execution of the ERCP technically difficult and some authors have reported a greater rate of complication due to the procedure [8,14,15] however recent series of cases report similar success rates in patients with and without PAD [16]. To our knowledge no study comparing ERCP with other approaches has ever been conducted, also data on the long-term benefits of the endoscopic technique are insufficient and limited to single centers series.

CONCLUSION

We would like to point out tree main highlights form these cases: 1) because the prevalence of paravaterian diverticula increases with age, it is possible to speculate that in an ageing population scenario such findings might come to the clinician’s attention more frequently than previously found; 2) it is possible that paravaterian diverticula cause cholangitis per-se, even without a direct evidence of bile duct calculosis (Lemmel’s syndrome), such was the case in the first patient described here; 3) because no evidence of benefit of any therapeutical approach over another has ever

been documented, and no guidelines addressing this clinical problem exist the therapeutical approach for patients with complicated paravaterian diverticula should be based on the clinical picture and evolution.

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