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Case Report

ENDOSCOPIC TREATMENT OF INTRALUMINAL DUODENAL DIVERTICULUM: A CASE REPORT

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SUMMARY – Intraluminal duodenal diverticulum is a rare entity that may cause recurrent pancreatitis. We report a case of a 36-year-old female admitted with etiologically unclear, recurrent pancreatitis. Radiographic study and endoscopy revealed a sac-like structure of the second portion of the duodenum near the minor duodenal papilla. Endoscopic diverticulotomy was successfully performed. No complications were observed and the patient was discharged home a day later. After 62-month follow-up, the patient was active and without symptoms. To our knowledge, this is the first reported case of recurrent pancreatitis caused by intraluminal duodenal diverticulum near the minor duodenal papilla, treated with endoscopic diverticulotomy performed from the apex to the base of the diverticulum.

Key words: *Endoscopic diverticulotomy; Intraluminal duodenal diverticulum; Recurrent pancreatitis; Duodenal minor papilla*

Introduction

Intraluminal duodenal diverticulum (IDD) is a rare congenital abnormality caused by failed recanalization of the occluded embryonic foregut. Diverticular attachment is most common on medial duodenal wall, proximal to the duodenal major papilla[1]. Histologically, IDD is covered on both sides by simple columnar epithelium and submucosa without muscular layer. Diverticulum usually measures 6-7 cm in length and 3-6 cm in width[1]. The entity was first described by Boyd in 1845[2]. However, the term "intraluminal duodenal diverticulum" was introduced by Kinzer in 1949[3]. It occurs equally in both genders but has a more variable time frame and nonspecific presentation such as abdominal pain, nausea, and vomiting. Reported complications also include perforation, fistula formation, partial or intermittent duodenal obstruction, gastrointestinal bleeding from ulcers, cholangitis, pancreatitis, and intestinal intussusception. There is a 40% incidence of coexistent abnormalities, including intestinal malrotation, annular pancreas, choledochocele, Hirschsprung's disease, imperforate anus, omphalocele, bladder exstrophy, hypoplastic kidneys, septate uterus, portal vein anomalies, polysplenia, and trisomy 21[1]. The 'windsock' sign is visual manifestation on upper gastrointestinal series. While surgery remains a cornerstone of treatment, the approach has seen innovations over the past few decades.

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Case Report

A 36-year-old woman previously of good health was admitted for abdominal pain. Laboratory findings confirmed acute pancreatitis (serum amylase 840 U/L, urinary amylase 2400 U/L, lipase 220 U/L, aspartate aminotransferase 25 U/L, alanine aminotransferase 28 U/L, bilirubin 19 mmol/L), for which she was hospitalized three months earlier, without clear etiology. Upper abdominal ultrasound and endoscopic ultrasound did not show any abnormality. The maximal bile duct diameter was 4 mm. The calcium value was normal. Contrast-enhanced abdominal computed tomography findings showed mild edema of the pancreatic head, which was in accordance with the diagnosis of mild acute pancreatitis. The possible causes of recurrent pancreatitis, mechanical (microlithiasis), toxic-metabolic (alcohol consumption, hypertriglyceridemia, hypercalcemia caused by hyperparathyroidism), anatomic (pancreas divisum), genetic (cystic fibrosis), autoimmune and occult neoplastic were all excluded. Magnetic resonance cholangiopancreatography was performed one month later and did not reveal any evidence for abnormal ductal dilatation and normal enhancement of pancreatic parenchyma. A sac-like structure of the second portion of the duodenum, measuring 7×2 cm, was seen. Upper gastrointestinal endoscopy showed intraluminal duodenal diverticulum (IDD) near the duodenal minor papilla, filled with food remnants, which was confirmed by upper gastrointestinal series with radiological contrast (Fig. 1). It was presumed that recurrent pancreatitis could be a consequence of compression of the minor papilla or distortion of the final part of the accessory pancreatic duct, caused by traction of the IDD by a trapped food bolus. Complete endoscopic diverticulotomy was performed in a tertiary endoscopic center in Belgium. First, filling of the IDD with radiographic contrast was done. It was followed by cutting the diverticulum wall from the outside in the direction from the apex to the base, first with a needle knife and then with an insulation-tipped endoscopic submucosal dissection knife. The diverticulum was thus fully opened and the walls were secured by clips. Cannulation of the minor papilla was performed to prevent periprocedural pancreatitis. Implantation of the stent was not successful because of highly elongated accessory pancreatic duct. Follow-up esophagogastroduodenoscopy was performed three months later to show complete healing and irregular elliptic opening of the duodenal second portion (Fig. 2). At 62-month follow up, the patient did not show any gastrointestinal related symptoms.

Discussion

As mentioned in previous reports, 20% of patients with IDD had acute or recurrent pancreatitis[1,4]. The pathophysiology is still unclear although Nance *et al*. hypothesize that it occurs due to obstruction of the papilla or to a change of the hydrostatic balance that promotes filling of the pancreaticobiliary duct with duodenal juice. The traditional standard treatment was surgery, in most cases duodenotomy or diverticulectomy[5]. Hajiro *et al*. first successfully performed endoscopic excision of the IDD in 1979[6]. We searched PubMed (December 2018) by using the terms "endoscopy" and "intraluminal duodenal diverticulum", and age (18+) and language (English and German) as filters. After exclusion of case reports describing surgical treatment and one paper written in Hungarian, we detected 23 papers with a total of 28 described cases. Law *et al*. have reported a total of five patients, which is the largest series of described patients in one paper[7]. Gender distribution of patients in the papers was equal, i.e., 14 male and 14 female patients. The mean age of patients was 43.57 years and median age 42.29 years. The most frequently reported symptoms were upper gastrointestinal symptoms (pain, nausea, vomiting). Five patients had pancreatitis, while another five patients had peptic ulcer and gastrointestinal bleeding. Only three patients had obstruction. Unlike Law *et al*., who performed endoscopic diverticulectomy in four of five patients described, the population of described patients was mostly treated with endoscopic diverticulotomy (16 of 28 patients)[7,8]. Restenosis and residual web were reported in only three cases[7-10]. In comparison with other reported cases, follow up in our patient was significantly longer. The advantages of endoscopic treatment, in comparison with surgery, are faster recovery and fewer postprocedural complications. Different endoscopic devices are used to perform these procedures. The use of a particular device should mostly depend on the gastroenterologist's experience. The presented overview of papers suggests us that the IDD should also be considered as a differential diagnosis in middle-aged patients with unspecific gastrointestinal symptoms. Radiographic imaging should always be performed before endoscopic procedures to exclude associated congenital abnormalities.

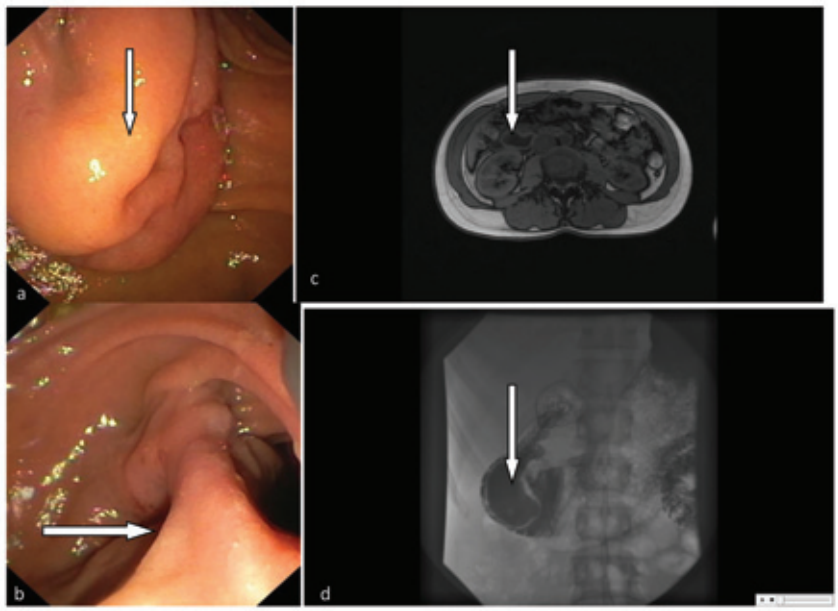

Fig. 1. (a) Intraduodenal diverticulum; (b) area of the duodenal minor papilla with orifice of intraduodenal diverticulum; (c) magnetic resonance cholangiopancreatography: a sac-like structure is seen in the second part of the duodenum; (d) display of intraduodenal diverticulum with radiological contrast which shows a blind-ended tubular structure from the medial segment of the descending duodenum (at the level of the papilla) to its inferior part.

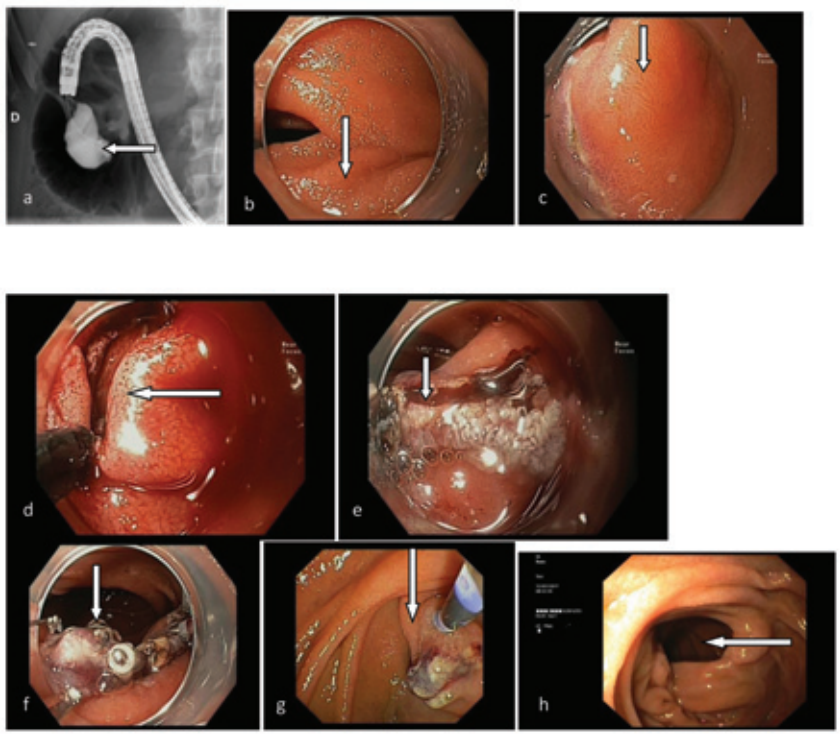

Fig. 2. (a) Display of intraduodenal diverticulum with radiological contrast at the beginning of endoscopic diverticulotomy; (b) and (c) endoscopic diverticulotomy at the beginning; (d) cutting intraduodenal diverticulum from the outside in the direction from the apex to the base; (e) fully opened intraduodenal diverticulum; (f) and (g) secured by clips and an attempt to implant stent in the minor papilla; (h) endoscopic finding 3 months later, entry into the second segment of the duodenum.

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Sažetak

ENDOSKOPSKO LIJEČENJE INTRALUMINALNOG DIVERTIKULA DUODENUMA: PRIKAZ SLUČAJA

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Intraluminalni divertikul dvanaesnika rijedak je, ali mogući uzrok recidivirajućeg pankreatitisa. U ovom prikazu slučaja radi se o 36-godišnjoj bolesnici zaprimljenoj zbog recidivirajućeg pankreatitisa nepoznate etiologije. Endoskopskom i radiološkom obradom verificirana je vrećasta struktura u drugom odsječku dvanaesnika, u neposrednoj blizini manje papile dvanesnika. Učinjena je endoskopska divertikulotomija uz uredan postproceduralni tijek te je bolesnica idućeg dana otpuštena kući. Nakon razdoblja praćenja od 62 mjeseca bolesnica je bez tegoba. Prema našim spoznajama, ovo je prvi opisani slučaj recidivirajućeg pankreatitisa uzrokovanog intraluminalnim divertikulom dvanaesnika u blizini manje papile dvanaesnika, gdje je endoskopska divertikulotomija učinjena od apeksa prema bazi divertikula.

Ključne riječi: *Endoskopska divertikulotomija; Intraluminalni divertikul dvanaesnika; Recidivirajući pankreatitis; Manja papila dvanaesnika*