AG-2019-171
dx.doi.org/10.1590/S0004-2803.202000000-19

Duodenal duplication cyst with enteroliths diagnosed by echoendoscopy and treated by endoscopic marsupialization

José Celso **ARDENGH**[1], Mariângela Ottoboni **BRUNALDI**[2], Rafael **KEMP**[3] and José Sebastião dos **SANTOS**[3]

Received: 10 December 2019
Accepted: 11 December 2019

Duodenal duplication cysts (DDC) are rare congenital anomalies primarily occurring during childhood. The duodenum is the site least frequently involved[(1)]. The symptoms of DDC depend on size and location; among adults, they frequently are of an obstructive nature and involve nausea, vomiting, pain, cholestasis, and pancreatitis. In the past, definitive diagnosis and treatment were usually based on a standard surgical approach[(2)]. DDC may contain stones, although this is a very rare presentation[(1,3-6)].

We report the case of a 31-year-old woman with an episode of epigastric pain of strong intensity 9 months ago, accompanied by nausea and vomiting, initially treated as dyspepsia. An abdominal ultrasound revealed cholecystolithiasis and a solid mass in the duodenum (5.0 x 4.9 x 2.6 cm) (FIGURE 1.A). Contrast examination of the duodenum revealed a filling defect occupying almost 90% of the intestinal lumen (FIGURE 1.B) and duodenoscopy revealed a subepithelial tumoral mass (FIGURE 1.C). The diagnosis of a DDC with an anechoic area filled with enteroliths was based on the findings of endoscopic ultrasound (EUS) (FIGURE 1.D).

Treatment consisted of endoscopic marsupialization during EUS using needle knife sphincterotomy in order to perforate the wall of the cyst and a polypectomy snare in order to expand the incision of the previous perforation[(*video)] (FIGURE 2.A). The lateral margins of the cyst were resected with a polypectomy snare and analyzed microscopically for confirmation of the DDC (FIGURE 3). Five months after the procedure, the patient was well, with no cyst recurrence (FIGURE 2.B).

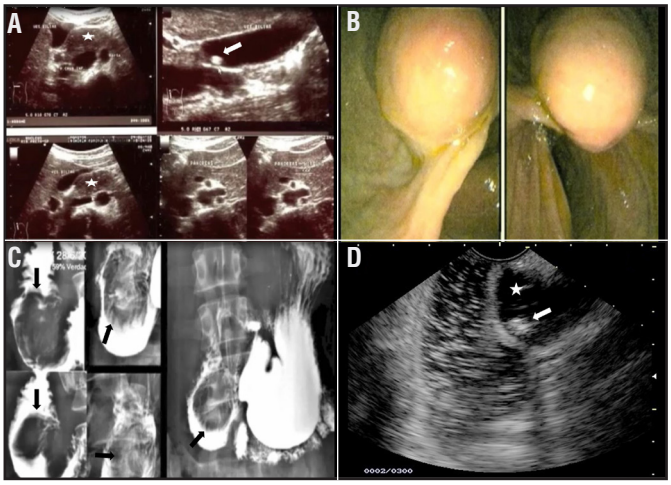

FIGURE 1. A- Abdominal ultrasound: duodenal cholecystolithiasis (arrow) and a nonspecific duodenal solid mass (*) measuring 5.0 x 4.9 cm. **B-** Endoscopic view: bulging formation with a normal mucosa close to the major duodenal papilla, feeling soft when touched with an endoscopic forceps (pillow sign). **C-** Contrast duodenography: filling defect (arrow) located close to the duodenal papilla and occupying almost 90% of the duodenal lumen. **D-** EUS: anechoic area (3.4 x 2.8 cm) (*) containing stones (arrow).

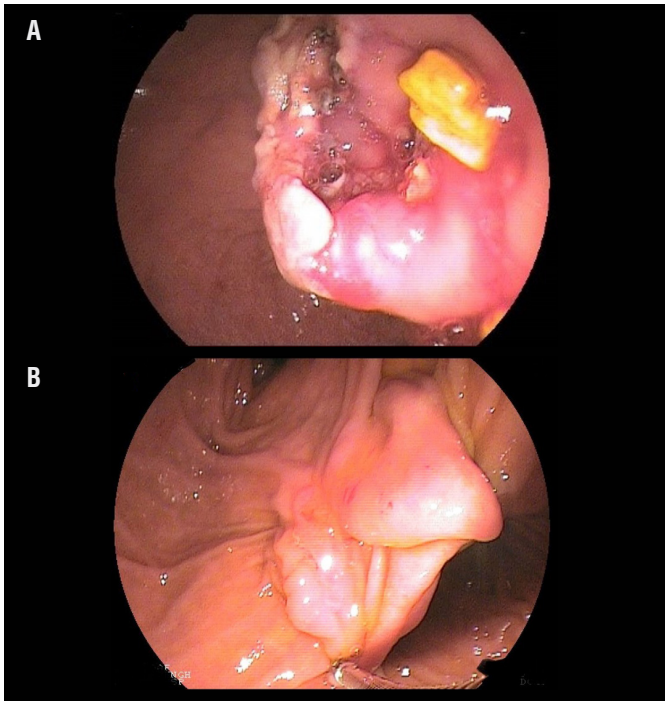

FIGURE 2. A- Endoscopic procedure: papillotomy performed in order to open the entire bulging area and to remove the stones. **B-** Control duodenoscopy five months after the procedure.

Declared conflict of interest of all authors: none
Disclosure of funding: no funding received
[1] Hospital 9 de Julho, Setor de Endoscopia, São Paulo, SP; Hospital das Clínicas da Faculdade de Medicina de Ribeirão Preto da Universidade de São Paulo (HCFMRP-USP), Setor de Endoscopia, Ribeirão Preto, SP, Brasil. [2] Universidade de São Paulo, Faculdade de Medicina de Ribeirão Preto, Departamento de Patologia e Medicina Legal; HCFMRP-USP, Ribeirão Preto, SP, Brasil. [3] Universidade de São Paulo, Faculdade de Medicina de Ribeirão Preto, Departamento de Cirurgia e Anatomia; HCFMRP-USP, Ribeirão Preto, SP, Brasil.
Corresponding author: José Celso Ardengh. E-mail: jcelso@uol.com.br
*** Video: https://www.youtube.com/watch?v=5tZgvgKcjFY**

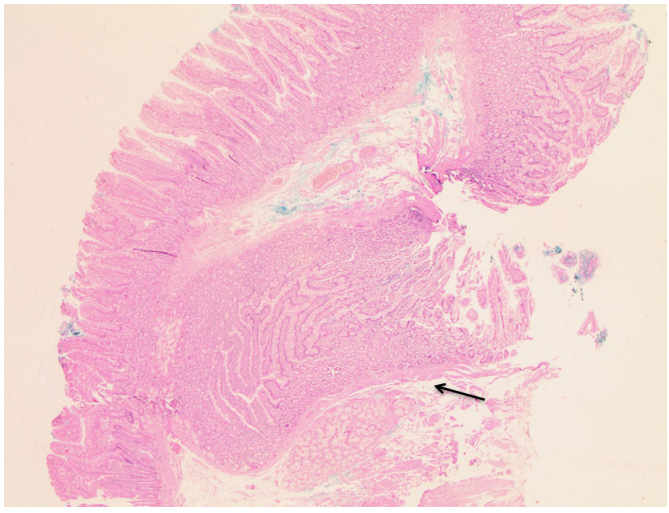

FIGURE 3. Histopathology: duodenal duplication cyst showing the invaginated duodenal wall in the submucosa (arrow). Panoramic view (original 20x magnification, hematoxylin and eosin).

DDC have a smooth muscle layer lined with intestinal mucosa, they communicate with some portion of the digestive tract, and are filled with clear fluid or bile when they are connected with the papilla. In addition, the presence of enteroliths may be attributed to food stasis and alkalinity[1,6].

In the present case, the diagnostic differentiation of the cystic mass observed had to be made between DDC and choledochal cyst. Taken together, EUS findings such as definition of the lesion's walls, the fluid content with enteroliths, the normal distal bile duct and the relationship with the major duodenal papilla were of help for the definition of the diagnosis and for the choice and execution of minimally invasive treatment by endoscopic marsupialization, with a tissue sample sufficient for histopathological diagnosis being obtained.

Malignant transformation of DDC is quite rare[7,8], but should be kept in mind in the decision-making process regarding partial or total resection. The elimination of secretion stasis in the residual lesion and repetition of endoscopic exams are recommended for patients submitted to endoscopic treatment[4].

Authors' contribution

Ardengh JC performed the procedure and wrote the paper. Kemp R helped with the procedure and with revision of the paper. Brunaldi MO carried out the histopathological study. Santos JS is the head of the service, supervised patient care and revised the paper.

Orcid

José Celso Ardengh: 0000-0002-5932-2499.
Mariângela Ottoboni Brunaldi: 0000-0002-7404-6779.
Rafael Kemp: 0000-0001-8008-2322.
José Sebastião dos Santos: 0000-0001-5118-0361.

Ardengh JC, Brunaldi MO, Kemp R, Santos JS. Cisto de duplicação duodenal com enterólitos diagnosticado por ecoendoscopia e tratado mediante marsupialização endoscópica. Arq Gastroenterol. 2020;57(1):110-11.

REFERENCES

1. Bar-Ziv J, Katz R, Nobel M, Antebi E. Duodenal duplication cyst with enteroliths: computed tomography and ultrasound diagnosis. Gastrointest Radiol. 1989;14:220-2.
2. Ildstad ST, Tollerud DJ, Weiss RG, Ryan DP, McGowan MA, Martin LW. Duplications of the alimentary tract. Clinical characteristics, preferred treatment, and associated malformations. Ann Surg. 1988;208:184-9.
3. Medina-Perez M, Garcia Ferris G, Caballero Gomez J. Recurrent acute pancreatitis caused by ampullar intestinal duplication filled with calculi. Rev Esp Enferm Dig. 1997;89:929-32.
4. Antaki N, Abboud D, Lemmers A, Antaki F, Deviere J. Acute recurrent pancreatitis secondary to the rare association of a duodenal duplication cyst and a pancreas divisum. Clin Res Hepatol Gastroenterol. 2013;37: e32-6.
5. Chryssostalis A, Ribiere O, Prat F. Endoscopic management of a duodenal duplication cyst filled with stones and revealed by recurrent pancreatitis. Clin Gastroenterol Hepatol. 2007;5:e31-2.
6. Leite AK, David AI, Szutan LA. Duodenal duplication cyst filled with enteroliths: a case report. Acta Gastroenterol Latinoam. 2009;39:125-8.
7. Falk GL, Young CJ, Parer J. Adenocarcinoma arising in a duodenal duplication cyst: a case report Aust N Z J Surg. 1991;61:551-3.
8. Inoue M, Nishimura O, Andachi H, Koga S. Early cancer of duodenal duplication. A case report. Gastroenterol JPN. 1979;14:233-7.

(cc) BY-NC