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CASE REPORT | PANCREAS

Giardia lamblia Infection in a Duodenal Duplication Cyst as a Potential Cause of Recurrent Acute Pancreatitis

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ABSTRACT

Giardia lamblia is a known etiological factor of a common infectious diarrhea. In rare cases, this parasite was found to be involved in the development of pancreatic and biliary tract diseases, both inflammatory and neoplastic. We present a case of a 64-year-old man known for a duodenal duplication cyst since 2013, with episodes of recurrent acute pancreatitis since 2017. He underwent endoscopic ultrasound-guided fine-needle aspiration of the duplication cyst, with evidence of *G. lamblia* infection. After treatment of giardiasis and evidence of parasite eradication, the episodes of recurrent acute pancreatitis ceased, following an ex adiuvantibus criterium.

KEYWORDS: acute pancreatitis; giardia lamblia; parasites; duodenal duplication cyst; endoscopic ultrasound

INTRODUCTION

Giardiasis is the most common waterborne parasitic infection of the human intestine worldwide, caused by the binucleated, flagellated protozoan parasite *Giardia lamblia*. Apart from the typical gastrointestinal clinical manifestations, many extraintestinal ones have been described over time. The pancreas and biliary tract are very unusual niches of the parasite, although a pathological involvement of these organs is possible.[1] We present a case of giardiasis accidentally diagnosed within a duodenal duplication cyst and likely conditioning recurrent acute pancreatitis (AP).

CASE REPORT

A 64-year-old man known for ischemic heart disease and type II diabetes mellitus since he was 45 years, in treatment with insulin and aspirin, with no family history of pancreatic diseases, living in the countryside, was accidentally diagnosed with a duplication cyst of the duodenal wall in 2013. He remained asymptomatic until 2017, when he developed 2 episodes of AP in January and May, respectively. Although the patient was known for asymptomatic gallstones, those episodes did not seem related to the biliary tract because during both hospitalizations, there was no evidence of biochemical alteration of liver function tests at the time of admission or throughout the hospitalization or radiological signs of cholelithiasis (aspartate transaminase 22 U/L, alanine transaminase 27 U/L, gamma glutamyl transferase 31 U/L, alkaline phosphatase 75 U/L, and total bilirubin 0.9 mg/dL in January; aspartate transaminase 27 U/L, alanine transaminase 35 U/L, gamma glutamyl transferase 36 U/L, alkaline phosphatase 81 U/L, and total bilirubin 1.1 mg/dL in May; immunoglobulin G4, calcium, and triglyceride levels were in the normal range; and no pancreatotoxic drugs were taken at both episodes). Magnetic resonance imaging with cholangiopancreatography at both admissions showed no other likely causes of the recurrent AP but confirmed the presence of the cyst (Figure 1). To better investigate the pancreatobiliary area and this cyst, he underwent a secretin-stimulation endoscopic ultrasound (EUS) (Figure 1). The examination showed a slightly heterogeneous echostructure of the pancreatic gland; a normal caliber and course of the main, dorsal, and ventral pancreatic ducts; and, in the context of the duodenal wall, a 14 mm peripapillary cyst creating a bulging in the second portion. This was slightly caudal compared with the major papilla and was characterized by all parietal layers on the sides of both the duodenal lumen and the pancreatic gland, highly

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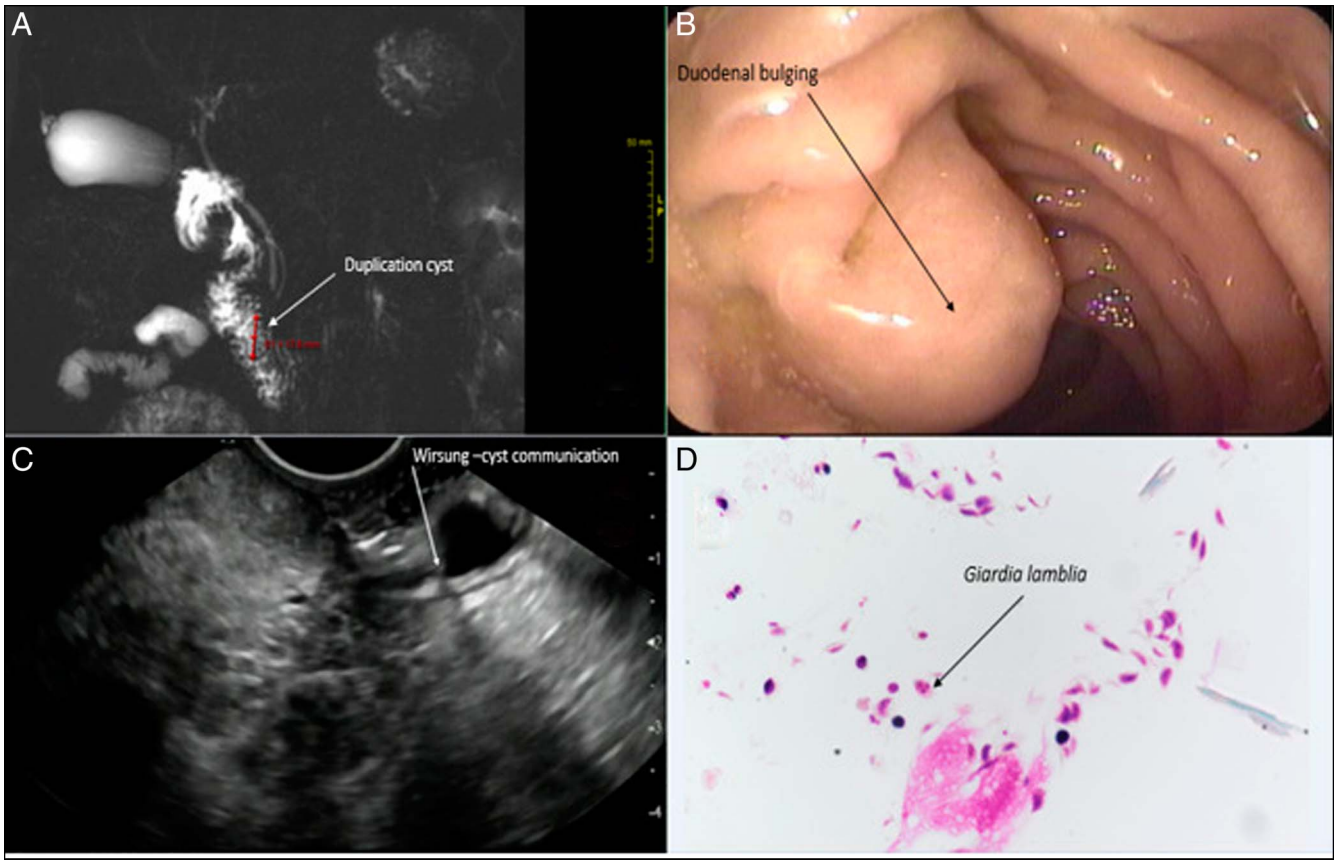

Figure 1. Duodenal duplication cyst infected with *Giardia lamblia* explored with different methods. (A) T2-weighted magnetic resonance imaging of the cyst next to the papillary area. (B) Endoscopic view of the duodenal bulging due to the cyst. (C) Endoscopic ultrasound image of the communication between the main pancreatic duct and the cyst. (D) Cytological smear of the duplication cyst liquid acquired through endoscopic ultrasound, with hematoxylin-eosin staining (600×), and evidence of multiple trophozoites attributable to *G. lamblia* infection (see the arrow for the typical *G. lamblia* shape).

suggestive for a duodenal duplication cyst. The use of secretin also highlighted a communication through a small duct (<1 mm in diameter) running within the duodenal wall between the cyst and the ventral pancreatic duct, not seen in the previous magnetic resonance imaging scan with cholangiopancreatography. The cystic fluid was then aspirated with a 22 gauge fine-needle aspiration needle, with complete cyst collapse and aspiration of 3 mL of a turbid colorless fluid from the cyst and with the presence of amylase and lipase. An unexpected finding was seen on the cytology smear: many pear-shaped protozoites in the context of mucus, compatible with *G. lamblia* (Figure 1).

The patient was treated with metronidazole, and eradication of the parasite was confirmed by both EUS-guided cyst aspiration, without the retrieval of parasites or mucus, and duodenal biopsies. After treatment, the man did not develop any further AP episode despite a follow-up of more than 4 years and the persistent presence of the 14 mm cyst at new magnetic resonance imaging.

DISCUSSION

Giardia lamblia is one of the most frequent parasites to infect humans. Most infected patients are asymptomatic, but infection may also manifest acutely as crampy abdominal pain with associated watery diarrhea or as a more chronic infection, mimicking irritable bowel syndrome or inducing malabsorption with weight loss. Rare case reports have also implicated the organism as a primary cause of biliary and pancreatic disease.

Miyahara et al[2] reported a case of giardiasis in a patient with pancreatic cysts and decreased pancreatic exocrine function, which was restored after treatment with metronidazole. Bathia et al[3] described a 16-year-old adolescent boy with X-linked hyperimmunoglobulin M syndrome who presented an increased alkaline phosphatase level (972 IU/L), biliary tree dilatation, distended gallbladder, and irregular bile duct wall with abrupt narrowing at the ampulla. Microscopic evaluation of the bile and of ampullary biopsy specimens showed multiple trophozoites and cysts of *G. lamblia*.

Giardiasis has been also described in association with a so-called pseudo-AP and with both pancreatic solid and cystic lesions.[4–7] In many cases, treating the infestation led to clinical resolution and disappearance of the cyst/lesion itself, although the pathophysiological link between the presence of the parasite and the development of these features is still unknown.[1,8,9] In our case, the onset of AP might be explained by different

hypotheses: (i) The biliary etiology is possible but less probable because of the normality of liver tests and radiological examinations during both admissions; (ii) the outflow of the pancreatic duct could be obstructed by the cyst growth, possibly stimulated by an inflammatory reaction to *G. lamblia* infection; and (iii) the parasite itself could be the cause of the pancreatic inflammation, and the presence of the duodenal cyst might be completely unrelated. These last 2 hypotheses are supported by the cessation of AP episodes after giardiasis eradication. Another scenario is that the cyst growth would still remain the primary cause of the pancreatic duct obstruction, being the presence of the duodenal parasite only a coincidence, yet the discontinuation of AP episodes could not be explained in any way. In this case, the complete clinical relief after *G. lamblia* eradication despite the reformation of the cyst leads to the parasite as the most likely direct cause of the recurrent pancreatitis. Generally speaking, the possibility of the EUS fine needle becoming contaminated by the *G. lamblia* passing through the wall of the small bowel exists, but in many cases, the description of the examined pathological sheets by multiple pathologists made the small bowel contamination less likely and aspirate from the pancreas more probable as a source of *G. lamblia*.[10] Other reports also retrieved the parasite on the pathological examination of the surgical pancreatic tumor specimen, with, therefore, no risk of contamination from the duodenum,[7] describing the parasite mostly in the pancreatic duct, with a possible ascending infestation of the gland.

In conclusion, in our case, it remains unclear in which step *G. lamblia* could have been involved, yet the giardiasis treatment interrupting the episodes of AP could follow an ex adiuvantibus criterium, making the *G. lamblia* the most probable cause of the recurrent AP.

DISCLOSURES

Author contributions: RPDL Pisani, G. Capurso, and L. Archibugi wrote the article. PG Arcidiacono, A. Laghi, and C. Doglioni contributed to the execution of the diagnostic and therapeutic management of the patient and to the ideation of the case and carefully reviewed the manuscript. L. Archibugi is the article guarantor.

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