

Letter to the Editor

SpyGlass percutaneous transhepatic cholangioscopy-guided diagnosis of adenocarcinoma of the ampullary region in a patient with bariatric biliopancreatic diversion

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To the Editor:

The definition of biliary stricture in patients with post-surgical biliary or enteric anatomy is challenging, due to limitations in accessing biliary system with a peroral approach. In these cases, percutaneous route is an option to obtain diagnosis and to steer patients' therapy [1]. However, cytologic findings obtained from bile and biliary brushing during percutaneous transhepatic cholangiography (PTC) demonstrated low sensitivity (43%) and high rate of inadequate specimens (21%) [2]. Nevertheless, cholangioscopy can be performed through percutaneous transhepatic access to obtain endoscopic visualization of the biliary tract and to guide tissue sampling with biopsy forceps [1,3]. Here, we report the diagnosis of adenocarcinoma of the ampullary region achieved at surgical specimen after endoscopic biopsy of dysplastic adenomatous alterations at the distal common bile duct obtained through percutaneous transhepatic cholangioscopy (PTCS) in a patient affected by worsening jaundice with previous bariatric biliopancreatic diversion.

A 61-year-old woman was referred to the Gastroenterology and Hepatology Unit of our hospital, with worsening jaundice for several weeks. She underwent bariatric biliopancreatic diversion and cholecystectomy 35 years ago. Her initial laboratory results showed elevated levels of bilirubin (total bilirubin: 27 mg/dL; direct bilirubin: 15 mg/dL), alkaline phosphatase (692 U/L), gamma-glutamyltransferase (172 U/L), aspartate aminotransferase (150 U/L) and alanine aminotransferase (103 U/L). Serologic tumor markers were normal, in particular carbohydrate antigen 19-9 (CA19-9, 7 UI/mL) and carcinoembryonic antigen (CEA, 3 UI/mL). Abdominal contrast-enhanced computed tomography (CECT) and magnetic resonance (MR) cholangiography demonstrated dilation of intrahepatic and extrahepatic bile ducts with stenosis of the distal common bile duct without intraductal stones or abnormalities of choledochal walls. Endoscopic retrograde cholangiopancreatog-

raphy (ERCP) and cholangioscopy with peroral approach were not feasible due to previous bariatric surgery and biliopancreatic diversion that hampered the achievement of the Vater papilla. On day 6 of hospital stay, a PTC confirmed the stricture of the distal common bile duct and an 8 Fr internal-external biliary pigtail drain (Flexima; Boston Scientific Inc., Natick, Massachusetts, USA) was positioned through right intrahepatic branch of the biliary tree which crossed the stenosis (Fig. 1A). No tumor cells were observed at cytologic examination of bile fluid and biliary brushing obtained during PTC. However, to define the etiology of the stenosis, a PTCS was performed. An informed consent was obtained from the patient. On day 12 of hospital stay, PTCS with SpyGlass DS (Boston Scientific Inc.) was performed under local anesthesia (carbocaine 2%) and conscious sedation (midazolam 5 mg) in the interventional radiology suite. The cholangioscope was inserted through an introducer of 11 Fr (Terumo, Tokyo, Japan) positioned on a dedicated guide of 0.035" (Jagwire; Boston Scientific Inc.) after removal of the 8 Fr percutaneous drain. A Gastroenterologist (Aragona G) with 17 years of experience maneuvered the cholangioscope, while an Interventional Radiologist (Michieletti E) with 21 years of experience in hepatobiliary procedures inserted and withdrew the cholangioscope under fluoroscopic guidance (Fig. 1B). The endoscopy documented a papillary exophytic lesion with superficial erosions of the distal common bile duct (Fig. 2A); the cholangioscope crossed the lesion, reaching the major papilla. Tissue samples were obtained by the Gastroenterologist with the SpyBite forceps (Boston Scientific Inc.). At the end of the procedure an 8 Fr percutaneous internal-external biliary drain (Flexima; Boston Scientific Inc.) was repositioned. The procedure was performed in 44 min, without periprocedural complications. The final pathologic examination suggested a diagnosis of choledochal villous adenomatous alterations with foci of dysplasia and stromal reaction (Fig. 2B). The patient was discharged on day 16 of hospital stay, with reduction of bilirubin (total bilirubin: 1 mg/dL; direct bilirubin: 0.3 mg/dL), alkaline phosphatase (287 U/L), gamma-glutamyltransferase (117 U/L), aspartate aminotransferase (95 U/L), and alanine aminotransferase (81 U/L). Thirty days after hospital admission, the patient underwent modified Whipple

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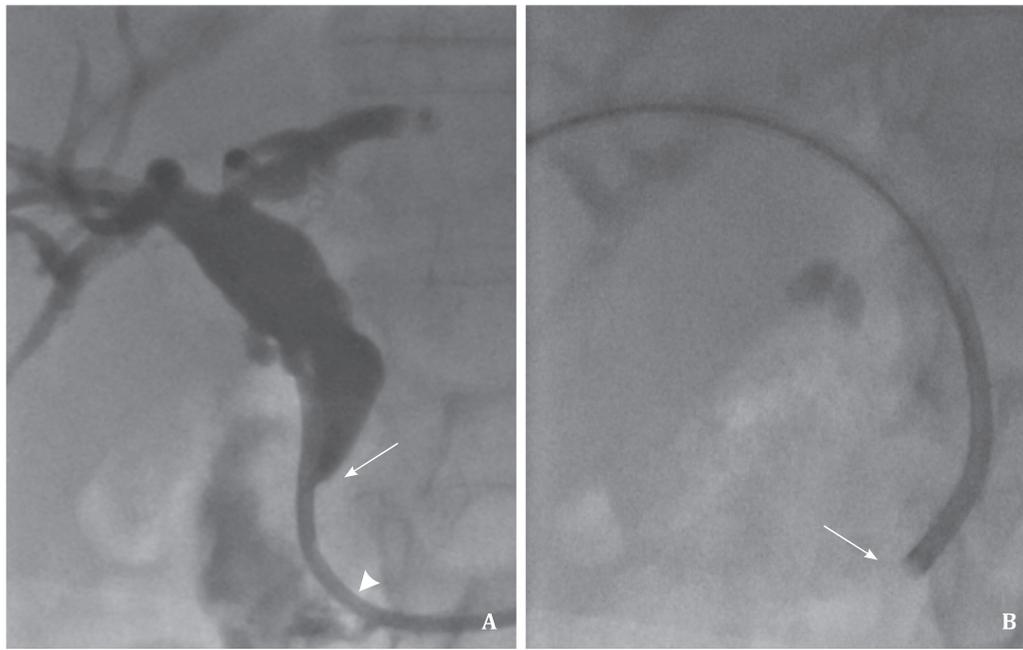


Fig. 1. A: Percutaneous transhepatic cholangiography (PTC) showed dilation of the intrahepatic and extrahepatic biliary tree, with stenosis (arrow) at the distal third of the common bile duct; during the procedure was positioned an 8 Fr internal-external biliary pigtail drain through right intrahepatic branch of the biliary tree which crossed the stenosis (arrowhead). **B:** During the percutaneous transhepatic cholangioscopy (PTCS) the interventional radiologist inserted and withdrew the cholangioscope visualizing it under fluoroscopic guidance (arrow), while the gastroenterologist maneuvered the cholangioscope and obtained tissue specimen of the lesion.

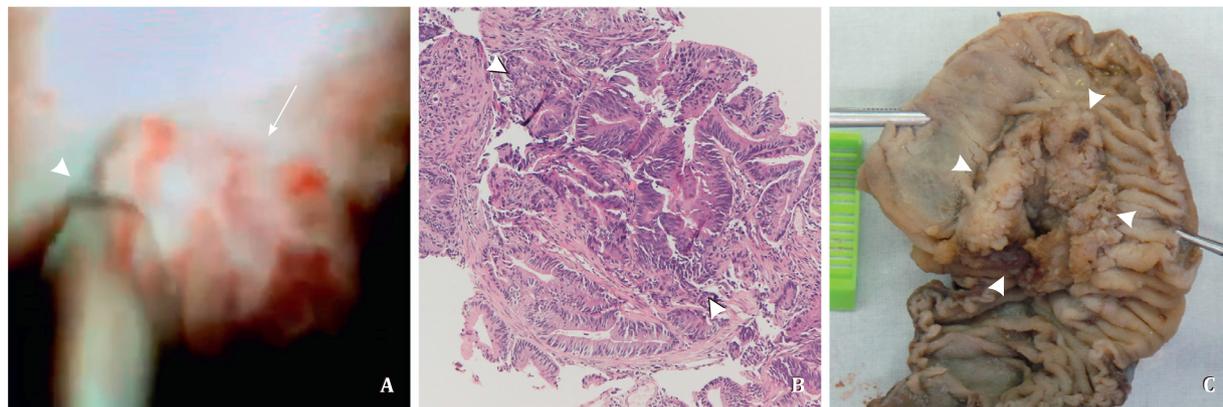


Fig. 2. A: Percutaneous transhepatic cholangioscopy (PTCS) performed with the Spyglass DS system showed the endoscopic appearance of a papillary exophytic lesion with superficial erosions at the distal common bile duct (arrow); the tissue specimen was obtained during the procedure with the SpyBite forceps (arrowhead). **B:** The biopsy obtained during PTCS documented a glandular epithelium in a villous disposition associated with foci of dysplasia (arrowheads; HE stain, $\times 100$). **C:** The gross examination of the surgical specimen showed a 4×2.5 cm vegetating mass protruding in the duodenal lumen (arrowheads) localized in the ampullary region.

procedure which consisted in resection of the distal common bile duct, of the pancreatic head, of the duodenum, and of the proximal 20-cm of jejunum, with creation of pancreaticojejunostomy and hepaticojejunostomy. The gross examination of the surgical specimen revealed a 4×2.5 cm soft tissue vegetating mass with superficial irregularity at the papilla of Vater, growing inside the duodenal lumen and obscuring ampullary orifice (Fig. 2C); the mass also involved the ampullary channel. At microscopic examination an intestinal type with partial mucinous aspects adenocarcinoma of the ampullary region was diagnosed, categorized as periampullary-duodenal subtype [4].

In previous studies, PTCS has been reported for lithotripsy of intrahepatic stone and for the diagnosis of indeterminate stricture of the biliary tree when cannulation of the Vater papilla during con-

ventional ERCP failed or was not feasible for post-surgical anatomy alterations of the biliary-intestinal anatomy [3,5,6]. In particular, a case series reported 4 patients with benign and malignant stricture of the biliary ducts diagnosed through PTCS performed with the first-generation fiberoptic system (SpyGlass; Boston Scientific Inc.), when conventional access to the biliary tree had not been possible [3]. In the case we presented, the diagnosis of choledochal dysplastic adenomatous alterations was achieved by the new digital system (SpyGlass DS), characterized by wider field of view, more stable image, and consistent orientation of the biopsy channel, in comparison to the first-generation system. At surgical specimen the final pathologic diagnosis was adenocarcinoma of the ampullary region. Using conventional ERCP, the new generation of Spyglass DS showed better sensitivity in diagnosis of biliary

malignancy (85% vs. 60%) and higher performance (adequate tissue specimen obtained in 97% of the patients vs. 88%) as compared to the first-generation cholangioscope [7,8]. The SpyGlass system is designed as a single operator tool for peroral cholangioscopy, however 2 operators are more adequate to use the cholangioscope in percutaneous transhepatic approach, as suggested in a previous report [3]. Particularly, the first operator performed the endoscopy and tissue acquisition with SpyBite forceps, while the second operator inserted and withdrew the catheter at the percutaneous entry port under visualization with fluoroscopy of the tip of the cholangioscope. The discrepancy between pathologic results obtained during PTCS and at surgical specimen could be explained by the characteristic of the periampullary-duodenal sub-type adenocarcinoma of the ampullary region. Typically, the pre-invasive neoplasm of the lesion corresponds to the vegetating component that grows along the duodenal wall and could extend intramucosally inside the intra-ampullary tract of the common bile duct; however, the invasive component is usually detected in the duodenal wall rather than inside the ampulla [4]. Since during PTCS only the ampullary component could be biopsied, the invasive component could not be documented.

In conclusion, when conventional access to the biliary tree is not possible with a peroral approach for post-surgical alterations of intestinal and biliary anatomy, PTCS with SpyGlass DS system is technically feasible and could achieve diagnosis of undetermined biliary ducts stricture.

Contributors

AG and ME proposed the study. CD, BFC, ZA, and MN performed the research and wrote the first draft. CD collected and analyzed the data. All authors contributed to the design and interpretation of the study and to further drafts. CD is the guarantor.

Funding

None.

Ethical approval

The patient agreed on the publication of her medical history and signed an informed consent.

Competing interest

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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Received 18 November 2018

Accepted 27 February 2019

Available online 5 March 2019