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Letter to the editor

Spyglass percutaneous transhepatic lithotripsy of symptomatic recurrent lithiasis of the intrahepatic bile duct with distal stenosis

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To the Editor,

Treatment of symptomatic intrahepatic bile duct stones is usually based on interdisciplinary plan (1). Endoscopic removal of intrahepatic stones through peroral or transhepatic route combined with lithotripsy are usually considered as first-line treatment, given the low complications rate (18% of the patients) and the high percentage of complete stone removal; percutaneous transhepatic cholangioscopic lithotripsy (PTCSL) shows better performance as compared to peroral approach (POCSL) for complete stone removal, obtained in 97% of the patients (vs 85% of POCSL) (2,3). Here, we report a case of recurrent unilateral symptomatic hepatolithiasis of the right posterior sectorial duct (RPSD) associated with biliary stricture, successfully treated by PTCSL and dilation with cutting balloon.

A 40-years old man was admitted to the Emergency Department for epigastric pain since several days in April, 2018. His laboratory findings showed an elevated total bilirubin level (2.7 mg/dl; normal values-NV-, 0-1.1 mg/dl), gamma-glutamyl transferase (GGT, 144 U/L; NV, 7-40 U/l), glutamic oxaloacetic transaminase (GOT, 45 U/L; NV, 10-37 U/L), glutamic pyruvic transaminase (GPT, 47 U/L; NV, 10-37 U/L), white blood cell count (WBC, 13.2x10^3/μl; NV, 4-10x10^3/μl) with absolute neutrophilia (10.3x10^3/μl; NV, 2-8x10^3/μl), and C-reactive protein (CRP, 5.1 mg/dl; NV, 0-0.5 mg/dl). The magnetic resonance (MR) cholangiography performed four days after admission identified hypointense filling defect representing a large stone of 2.3 cm in the RPSD causing a dilation of the afferent segmental ducts. Ten days after admission, the patient underwent percutaneous cholangiography that confirmed the obstruction of the RPSD sustained by the stone and an 8Fr internal-external drain was positioned. At 16th day of hospitalization, the patient underwent a combined percutaneous and endoscopic retrograde cholangiopancreatography (ERCP) procedure with the "rendez vous" technique performed by Interventional Radiologist and Gastroenterologist. Using a 11.5 mm Standard Occlusion Balloon (Boston Scientific Inc., Natick, Massachusetts, USA) the stones were pushed in the common bile duct (CBD) and in duodenum following the papillary sphincterotomy provided by ERCP. Furthermore, at the final radiogram, was displayed a stricture of the RPSD, treated by cholangioplasty (6x20 mm Peripheral Cutting Balloon, Boston Scientific Inc.). At the end of the procedure remained a partial stricture and an 8Fr internal-external drain was left, substituted 13 days later by a 12Fr plastic prosthesis (Gran Torino; Meditalia Biomedica, Genova, Italy), removed by ERCP two months later. The patient was dismissed at home one-month after admission, without symptoms and with normalization of bilirubin level (0.2 mg/dl), GGT (39 U/L), GOT (18 U/L), GPT

(22 U/L), WBC (8.2x10^3/μl), and CRP (0.4 mg/dl). Seventeen months after the first hospital admission, in September 2019, the patient manifested fever, epigastric pain and vomiting. At casualty laboratory findings showed increased levels of bilirubin (2.8 mg/dl), GGT (98 U/L), GOT (594 U/L), GPT (777 U/L), WBC (11.2 x10^3/μl), and CRP (1.5 mg/dl). The cholangiography performed one-day after admission, displayed dilation of the posterior sectorial intrahepatic biliary tree with three recurrent stones of the RPSD with overall 2.2 cm diameter (Figure.1A); after crossing the stricture, an 8Fr internal-external drain was positioned. ERCP failed to directly visualize the stone and after the failure of several combined (percutaneous associated with ERCP) attempts to sweep stone in the common bile duct (CBD), was decided to proceed to PTCSL (Video) with the SpyGlass Direct Visualization System II (SDVS II; Boston Scientific Inc.). Four days after admission PTCS was performed under local anesthesia (carbocaine 2%) and conscious sedation (midazolam 5 mg) in the interventional radiology suite. The patient received prophylactic ciprofloxacin 400 mg intravenously the day of the procedure. The 8Fr percutaneous drain was substituted by an introducer of 11Fr (Terumo, Tokyo, Japan). The SpyScope with an outer diameter of 10Fr was inserted through the introducer on a dedicated guide of 0.035" (Jagwire; Boston Scientific Inc.). The SpyScope provides four-way steering of the tip and accommodates four channels: two for water irrigation, one for the fiber-optic device (6000 pixel image with 70° field of view) and one for application of accessory devices such as a biopsy forceps (SpyBite, single-use, 1.0 mm diameter with 4.1 mm jaw opening at 55°) or a 1.9Fr probe for the electrohydraulic lithotripsy (EHL) connected with the Autolith system (Northgate Technologies, Elgin, Illinois, USA). A Gastroenterologist (X.X) with 18 years of experience maneuvered the cholangioscope, while an Interventional Radiologist (Y.Y) with 22 years of experience in hepatobiliary procedures inserted and withdrew the SpyScope under fluoroscopic guidance. The endoscopy identified several stones in the RPSD. Afterwards, stones were fragmented with EHL and the fragments were pushed in the CBD with 11.5 mm Standard Occlusion Balloon (Boston Scientific Inc.). The PTCS showed a stricture of RPSD confluence with the main right hepatic biliary duct, sampled through the SpyBite forceps (Boston Scientific Inc.), with the histologic diagnosis of fibrosis. Stricture cholangioplasty with 6x20 mm cutting balloon (Peripheral Cutting Balloon, Boston Scientific Inc.) was performed, with improved stenosis and no intrahepatic stones (Figure. 1B). At the end of the procedure, an internal-external 8Fr drain was left. Three days later, after removing internal-external drain, two 12Fr plastic prosthesis (Gran Torino; Meditalia Biomedica, Genova, Italy) were deployed crossing residual stenosis. The patient was dismissed 14 days after admission, asymptomatic, with normalization of bilirubin level (0.6 mg/dl), GOT (29 U/L), WBC (7x10^3/μl) and reduction of GGT (65 U/L), GPT (84 U/L), and CRP (0.8 mg/dl). Four months later the plastic prosthesis were removed. Fifteen months after lithotripsy, in December 2020, the patient remained asymptomatic, and the MR cholangiography (Figure. 2) showed a marginal dilation of the right intrahepatic posterior duct sustained by a residual, minimal, stenosis of the RPSD, however without signs of intrahepatic biliary lithiasis.

POCSL and PTCSL are indicated for patients with indeterminate biliary strictures (when their nature cannot be determined after basic laboratory work-up, abdominal imaging and ERCP) or with difficult bile stones (challenging access route, size ≥15 mm, number, impaction, intrahepatic location or the presence above a stricture) (4). PTCSL with SDVS shows slightly higher procedural success as compared to POCSL for complete stone removal (97% vs 85% of the procedures) and lower complications (12% vs 21% of the cases) (2, 3). When ERCP or POCSL failed to remove intrahepatic biliary stones sustained by downstream biliary stricture, as in the present case, further attempt with percutaneous route are considered. In comparison to POCSL, PTCSL is traditionally limited by fistula dilatation and maturation which can require around two weeks (4). This time is longer for larger cholangioscopes, with outer diameter of 4.9–5.2mm, that require an introducer of 16-18Fr and 10–12 days for fistula dilatation and maturation time (4). SDVS II has an outer diameter of 3.3mm (10Fr) that can be accommodated by an introducer of 11–12Fr (5). The fistula dilatation and maturation time for SDVS II is not well defined in literature (5).

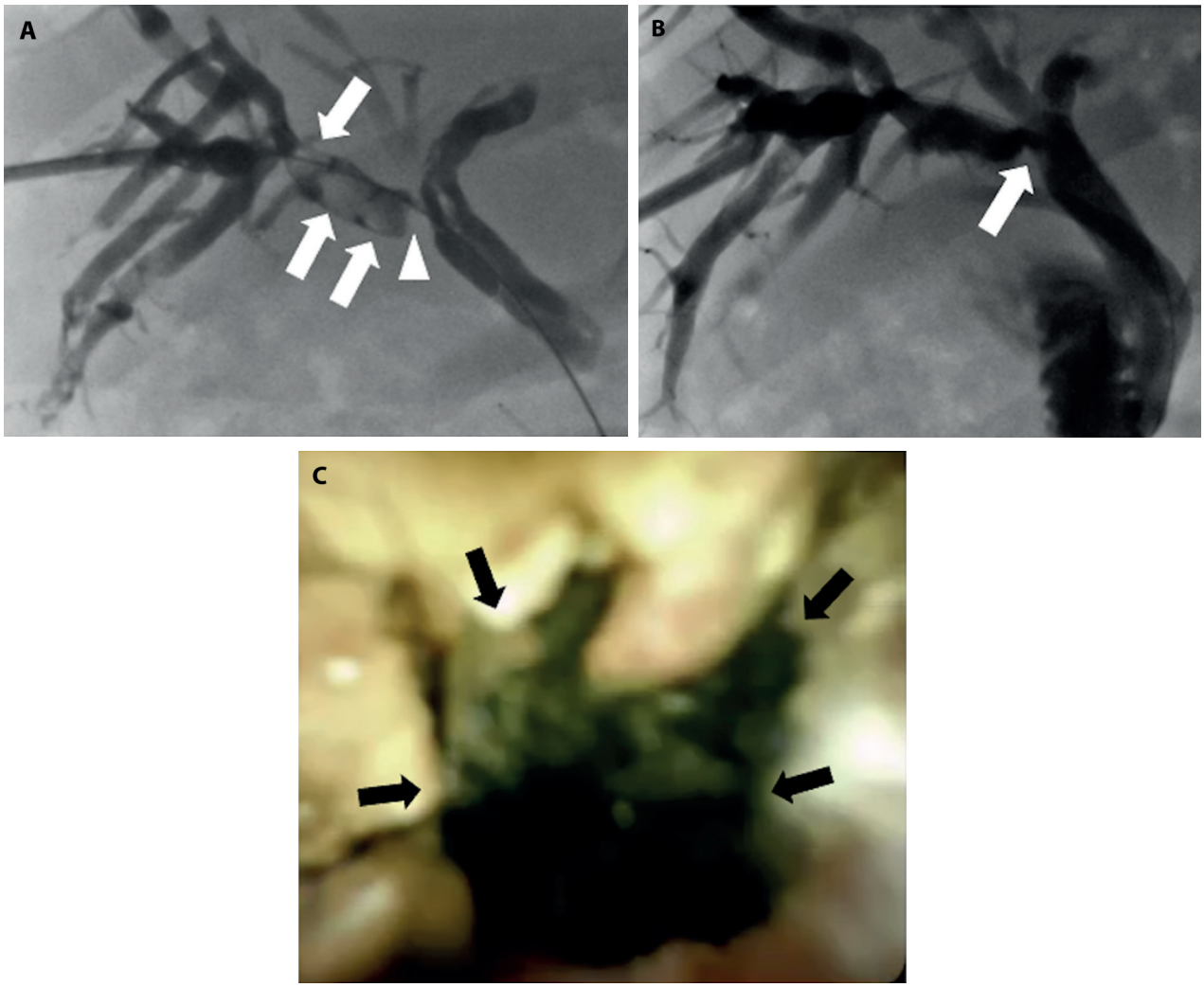

Figure 1. (A) Percutaneous transhepatic cholangiography showed three filling defect representing biliary stone in the right posterior intrahepatic bile duct (arrows) and a stricture at the confluence of the right posterior duct in the main right hepatic duct (arrowhead). (B) After percutaneous transhepatic cholangioscopic lithotripsy and cholangioplasty with cutting balloon remained a moderate stenosis at the confluence of the right posterior duct in the main right hepatic duct (arrow) without residual stones. (C) The endoscopic visualization of the biliary stone (arrows) in the right posterior hepatic duct during the percutaneous transhepatic cholangioscopy provided by the SpyGlass Direct Visualization System II (Boston Scientific Inc., Natick, Massachusetts, USA).

In the present case, the 8Fr biliary drain was left for three days, and at the beginning of the procedure the percutaneous tract was upsized to accommodate an 11Fr introducer, with 1mm dilatation of the tract diameter. As compared to larger cholangioscopes, SDVS II can reduce the traumatism required for fistula dilatation and the related complications, which are more frequent during this procedural phase (around 13% of the procedures) as compared to tract maturation or therapeutic percutaneous transhepatic cholangioscopy (7% for both phases) (4). The use of holmium:yttrium-aluminum-garnet achieved an higher success rate as compared to EHL (100% vs 97%) for the treatment of intrahepatic biliary calculi (2, 6). However, in the present case EHL was employed considering its lower setup cost and wide availability; furthermore PTCSL with EHL remains an effective (success rate of 97%) and safe procedure (complications occurrence around 12%) with low recurrence rate (10%) (2). As in the present case, recurrence rate of intrahepatic lithiasis

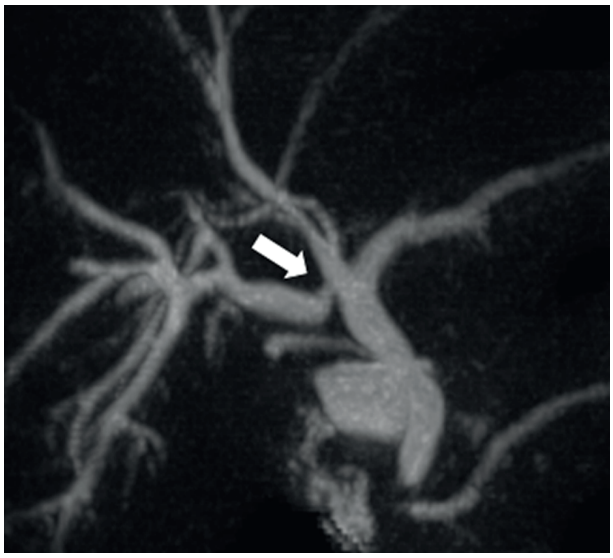

Figure 2. The three-dimensional T2-weighted Turbo Spin Echo SPACE image performed during magnetic resonance-cholangiography performed fifteen months after percutaneous transhepatic stone lithotripsy, showed no changes of the stricture at the confluence of the right posterior with the main right hepatic bile duct (arrow), without relapsing stones.

is high in patients with biliary strictures, reported in around 60% of the patients with a median recurrence time of 11 months; for this reason treatment of biliary stricture is paramount (7). The treatment of biliary strictures relies on several endoscopic techniques, including dilation with conventional or cutting balloons (8-10). In particular, late procedural success rate is lower with conventional balloon (82% at a mean time of 30 months) in comparison to cutting balloons (100% at a mean time of 14 months) for biliary stenosis in biliary-enteric anastomotic strictures or after liver transplantation (8, 9). The stricture of intrahepatic duct treated in the present case was approached with cutting balloon without recurrence after 15 months. In our knowledge this is the first case of intrahepatic ducts treated by cutting balloon.

In conclusion, PTCSL with EHL performed with SpyGlass Direct Visualization System II enables to resolve recurrent intrahepatic stones caused by benign biliary stricture of the right posterior intrahepatic duct; the combined treatment of the stones with PTCSL and of the stricture by cutting balloon dilation achieved early technical success and no recurrence after 15 months.

Credit authorship contribution statement: Colombi D, Morelli N: Data curation, Formal analysis, Investigation, Methodology, Project administration, Validation, Visualization, Writing - original draft. Colombi D, Aragona G, Michieletti E: Conceptualization, Supervision, Writing - review & editing. All authors have approved the final article.

Ethical approval: since this case is retrospective, the ethical approval was waived by our Institution.

Conflicts of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

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