Biliary pneumatosis: a new finding in a patient with cholangitis

A 72-year-old man with a previous history of renal transplantation for autosomal dominant polycystic kidney disease and secondary sclerosing cholangitis caused by recurrent bile duct injury with hepatolithiasis was admitted with a new episode of cholangitis, with a gallstone, but no signs of septic shock. Additionally, he had undergone cholecystectomy and Roux-en-Y hepaticojejunostomy 2 years previously.

During the current hospital admission, percutaneous transhepatic cholangiography showed multiple strictures along the right and left intrahepatic ducts, with a gallstone in the left duct. Percutaneous radiological management failed because of an impassable stenosis. Thereafter, endoscopic duodenojejunostomy with a lumen-apposing metal stent (LAMS; Hot-AXIOS; Boston Scientific Co., Marlborough, Massachusetts, USA) was performed to facilitate access to the afferent limb and complete an evaluation of the left intrahepatic duct.

A gastroscope was introduced into the hepaticojejunostomy through the LAMS (▶Video 1). A calculus within segment III of the liver was observed and fragmented using electrohydraulic lithotripsy (▶Fig. 1 and ▶Fig. 2a). Additionally, the endoscopy revealed submucosal blebs in the left intrahepatic bile duct (▶Fig. 2b, c; ▶Video 1), resulting in benign bile duct strictures. Some of the blebs were punctured with a sclerotherapy needle to confirm the presence of gas in the hepatic duct walls (▶Fig. 2d). The appearance of the gas-filled blebs was consistent with biliary pneumatosis. The patient was discharged home on day 3 after his endoscopy.

This novel finding had not been observed on other previous imaging tests and persisted in the following endoscopic examinations. No additional signs of pneumatosis intestinalis were seen. Given the mechanisms of pneumatosis cystoides intestinalis, either mechanical damage due to previous radiological interventions or a bacterial etiology in the setting of multiple episodes of cholangitis could explain the submucosal bleb formation [1–3]. Finally, considering the evolution of advanced endoscopy for biliary access and endoscopic exploration of the biliary ducts, this could be the first description of many other cases.

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Competing interests

Dr. Aparicio is a consultant for Boston Scientific. The remaining authors declare that they have no conflict of interest.
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Fig. 2 Endoscopic views showing: a hepatolithiasis fragmentation using electrohydraulic lithotripsy; b the hepatic duct bifurcation with a gas-filled bleb; c another submucosal bleb within the left intrahepatic duct; d puncture of a bleb with a sclerotherapy needle releasing gas.