Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.ciresp.2022.10.021.

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High-grade dysplasia in the cystic duct after cholecystectomy

Displasia de alto grado en el conducto cístico tras colecistectomía

Laparoscopic cholecystectomy is one of the most frequently performed surgeries. High-grade dysplasia (HGD) in the cystic duct resection margin is uncommon after cholecystectomy for cholelithiasis (<0.1%) or acute cholecystitis (1%), and it is considered a precursor of gallbladder and bile duct cancer.[1,2] Therefore, when there is a finding of dysplasia, it is necessary to rule out concomitant tumor pathology, mainly multifocal biliary tumors (biliary intraductal papillary neoplasm, gallbladder adenocarcinoma and cholangiocarcinoma).[2] However, when faced with an isolated HGD with no clear signs of invasion, there is no therapeutic consensus.[3]

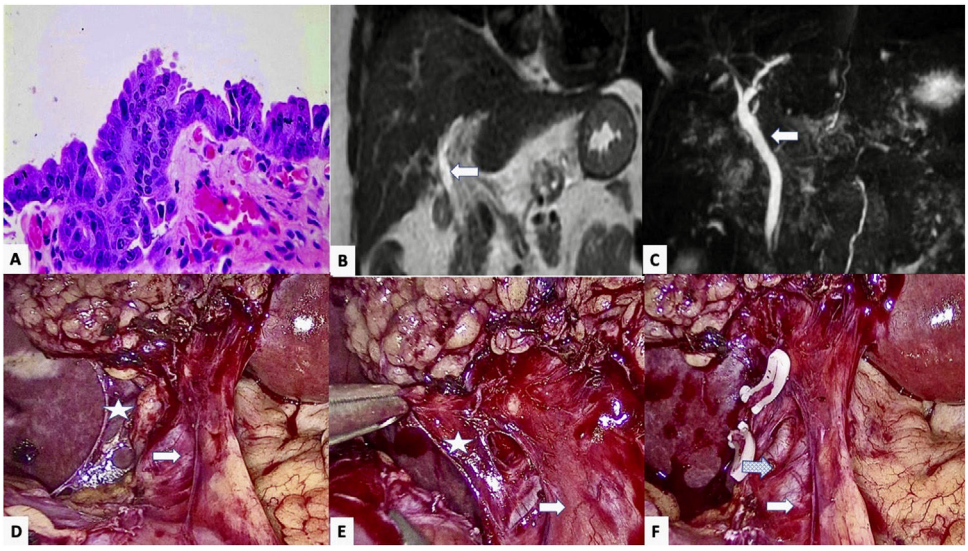

Fig. 1 – High-grade epithelial dysplasia in the cystic duct resection margin of the cholecystectomy specimen, showing loss of nuclear polarity and intense atypia with nuclear hyperchromatism, irregularity and enlargement; B and C) Main bile duct (Arrow) in coronal MRI slices in T1 (B) and T2 (C), with no observed stenosis or dilatation; D and E) Laparoscopic view, showing the main bile duct (arrow) and the stump of the cystic duct (Star) prior to its surgical resection; F) Image after resection of the cystic remnant, showing the main bile duct (arrow) with associated varicose vein (dotted arrow).

We present a 44-year-old woman with a medical history of multiple sclerosis, bone marrow transplant for acute myeloid leukemia, and cholelithiasis with various episodes of biliary colic over the course of a year, for which she underwent elective laparoscopic cholecystectomy (performed without incident). The histopathological study revealed a 2-mm high-grade dysplasia (high-grade biliary intraepithelial neoplasm) in the resection margin of the cystic duct, yet no involvement of the gallbladder (Fig. 1A). On physical examination, the patient presented no abdominal pain or jaundice, nor did she report weight loss. The blood work-up was normal, showing no changes in liver enzymes or bilirubin, and tumor marker levels were normal (CEA 3.09 ng/mL, Ca 19.9 18.80 U/mL). An MR cholangiopancreatography (MRCP) of the liver detected no space-occupying lesion (SOL), pathological lymphadenopathies, or dilation of the bile duct (Fig. 1B and C).

Based on the histology report and the absence of distant disease, the multidisciplinary committee ruled out any further diagnostic studies (CT, ERCP) and proposed a surgical intervention for the re-resection of the cystic margin. The procedure was laparoscopic and entailed dissection and division of the cystic remnant, proximal to the bile duct (Fig. 1D and F). The intraoperative study of the resection margin ($1 \times 0.6 \times 0.1$ cm) as well as the re-resection of the cystic gland showed no residual dysplasia or malignancy. The postoperative period was uneventful. One month after surgery, the follow-up (MRCP) revealed nothing of interest.

The appearance of HGD in the cystic margin after cholecystectomy is a rare finding (0.2%–1%).[3,4] Its importance lies in the potential progression to carcinoma (cholangiocarcinoma in the bile duct or gallbladder adenocarcinoma), as it is a common precursor (69% of cases) and both have an ominous prognosis with invasion.[3,4] If the diagnosis of malignancy is confirmed, surgical treatment must be aggressive and include: bile duct resection; hepatectomy of segments IVb and V; and lymph node dissection of the portal, perihilar, and gastrohepatic ligament nodes.[1] Some groups have described a correlation of HGD with extrahepatic cholangiocarcinoma (13%–20%) and also report that that the main prognostic factor for survival was surgical margin involvement.[4–6] Due to the multifocal nature of tumors in this location, exploration of the bile duct is mandatory, requiring resection in many cases (Table 1).[7]

However, the presence of HGD when there is no suspected malignancy is quite rare (0.05%).[8,9] In these patients, the main objective, as highlighted by the guidelines of the American Hepato-Pancreato-Biliary Association (AHPBA), is to rule out adjacent tumor pathology,[10] initially based on intraoperative findings of the previous cholecystectomy, presence of lymphadenopathies, and dilation of the bile duct, gallbladder or biliary tumor. Secondly, after the histological confirmation of HGD, an extension study is necessary with abdominal CT and MRCP. Tumor markers (CEA, CA 19.9) could help guide the diagnosis. As previously mentioned, this type of neoplasm tends to be multifocal, so a diagnostic ERCP could be appropriate.

Preoperative diagnostic tests frequently do not provide relevant information. Thus, the question that remains is whether radical surgery is necessary or whether cystic resection and close surveillance would suffice in the

Table 1 – Distribution of the HGD cases described in the literature.

Cases	Age (years)	Sex	Preoperative image	Surgery	Histology	Survival
Bickenbach KA et all, 2011. (N = 5)	65.2 (57–77)	Males (3:5)	80% MRI 40% CT + MRI 20% ERCP + MRI	Resection of cystic +/– (80%) lymphadenectomy +/– (40%) segmentectomy IV-V	80% no dysplasia 20% cholangiocarcinoma (T2N1)	80% disease-free 20% death
Sunkel-Laing B et al, 2014. (N = 1)	67	Male	CT + ERCP	Pancreaticoduodenectomy (Whipple)	Cholangiocarcinoma	Death
Moslim MA et al, 2017. (N = 1)	36	Female	MRI + ERCP + CT	Cystic resection	No dysplasia	Disease-free
Saliba M et al, 2019. (N = 1)	44	Male	MRI	Cystic resection	No dysplasia	Disease-free

absence of diagnosis or evidence of malignancy. In this context, the role of multidisciplinary committees becomes essential. Until now, radical surgery with excision of the bile duct has been considered the appropriate therapy. However, the most recent published papers advocate a conservative approach with close monitoring, either with or without surgical treatment.[1,4] Due to its importance, this decision must always be made with the patient, most whom opt for surgical treatment, as described in other series.[4] The diagnosis is histological, based on the detection of cells with HGD, suggesting that the disease-free resection margin should be at least 5 mm, in prognostic terms.

In our case, we decided to reoperate with minimally invasive surgery, resecting the cystic remnant and confirming the absence of margin involvement with an intraoperative sample. This latter technique is not mandatory due to its low diagnostic yield (fibrosis, inflammatory changes), but it helps determine the prognosis and extension of surgery.[3] Regarding postoperative follow-up, the data in the literature are ambiguous. Nevertheless, a follow-up of at least 5 years is recommended, similar to biliary carcinoma, with tumor markers (CEA, CA 19.9) and abdominal ultrasound/CT scan every 6 months for 2 years, and annually thereafter.

In conclusion, HGD of the cystic remnant with no evidence of malignancy after cholecystectomy is a rare pathology. However, it is necessary to rule out its association with biliary tumor pathology, and the treatment of choice includes cystic resection and close postoperative follow-up.

Conflicts of interest

None of the authors have any conflicts of interests.

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Sump syndrome as a late complication of side-to-side hepaticojejunostomy due to biliary iatrogenesis

El síndrome de sumidero como complicación tardía de la hepaticoyeyunostomía latero-lateral por yatrogenia biliar

Sump syndrome (SS) is defined as the accumulation of bile, biliary sludge, calculi or detritus in the bile reservoir (infraanastomotic common bile duct), either after side-to-side choledochoduodenostomy as the most frequent cause,[1,2] or after side-to-side Roux-en-Y hepaticojejunostomy.[3]

We present 2 cases of SS. The first is a 74-year-old man who underwent Roux-en-Y side-to-side hepaticojejunostomy 26 years ago due to iatrogenic injury to the hepatic duct during laparoscopic cholecystectomy. In the past 2.5 years, he had been hospitalized for multiple episodes of acute cholangitis and various liver abscesses. The patient was treated with antibiotic therapy, PTC-guided dilatations of the hepaticojejunostomy, and placement of a biodegradable stent in 2019. MRI was not performed due to the presence of shot (pellets) after a hunting accident; instead, a CT scan showed dilation of the intrahepatic and extrahepatic bile ducts occupied by gallstones. After a failed papillotomy to remove the common bile duct stones, the patient came to our hospital, where surgery was indicated. Surgery performed 4 months ago revealed almost complete obstruction of the side-to-side hepaticojejunostomy as well as the intra- and extrahepatic bile ducts dilated with biliary sludge and abundant soft calculi (Fig. 1a). A 45-cm afferent jejunal loop was observed; the hepaticojejunostomy was divided, and stones and sludge were removed from the intra- and extrahepatic bile duct using lavage and a Fogarty catheter. Clear passage to the duodenum was confirmed, and the common bile duct was sutured. The bile duct at the confluence was widened to 2.5 cm in diameter by longitudinal division of the left hepatic duct, concluding with an end-to-side hepaticojejunostomy and repositioning of the afferent loop 70 cm from the biliojejunal anastomosis. The postoperative period was uneventful, and the patient has been asymptomatic ever since.

The second case is a 68-year-old male patient with a history of a Roux-en-Y side-to-side hepaticojejunostomy after choledochal stenosis secondary to Kehr tube placement. Four years later, he presented an episode of acute cholangitis; MRI detected lithiasis and a 1.5 cm dilation of the intra and extrahepatic bile duct on magnetic resonance cholangiopancreatography (MRCP) (Fig. 1b). Surgery was indicated due to intrahepatic lithiasis and SS, and we confirmed the permeability of the hepaticojejunostomy (1 cm diameter), intrahepatic and extrahepatic biliary dilatation (1.8–2 cm), and abundant intrahepatic and extrahepatic calculi. As in the first case, the bile duct was enlarged to 3 cm at the confluence, extracting intrahepatic and common bile duct stones and subsequently performing end-to-side Roux-en-Y hepaticojejunostomy with the biliopancreatic limb at a distance of 70 cm. After an uneventful postoperative period, the patient remains asymptomatic 3 years later.

In spite of its rarity, the most frequent presentation of SS is 5–9 years after side-to-side choledochoduodenostomy. Symptoms at presentation may include abdominal pain, fever, cholangitis, pancreatitis, liver abscesses, and moderately elevated liver enzymes, while ERCP sphincterotomy is currently the accepted treatment of choice.[2,4,5] When endoscopic treatment is not possible or insufficient, surgical intervention is the most accepted indication.[1,2,5]

Risk factors for SS are long common bile duct length (below the choledochoduodenostomy or hepaticojejunostomy), choledochoduodenal or hepaticojejunal stenosis, residual or recurrent choledocholithiasis, and papillary stenosis or dysfunction.[2,6,7] A case similar to ours is the Kim et al. report[3] (Table 1) of a patient treated with side-to-side hepaticojejunostomy who, 26 years later, debuted with symptoms of leukocytosis and a liver abscess measuring 6.5 cm, which was treated with antibiotic therapy, percutaneous drainage and ERCP/papillotomy, extracting calculi and sludge from the common bile duct and showing no evidence of hepaticojejunal stenosis. The indication for surgery in our first case was due to almost complete obstruction of the hepaticojejunostomy and intrahepatic and extrahepatic lithiasis with several failed attempts at