An Unexpected Case of Obstructive Jaundice: Biliary IPMN

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Case Report

A man with a previous gastric GIST resection and total proctocolectomy in ulcerative colitis presented to our center for a first episode of obstructive jaundice due to a tubular adenoma of the papilla, and was treated with an endoscopic resection and biliary stent placement. In December 2015, the patient presented with acute cholangitis; he underwent several ERCPs with multiple biliary stenting procedures, as well as standard radial EUS and several other ancillary techniques: Intra Ductal Ultra Sound (IDUS), ERCP with cholangioscopy and intraductal biopsies, probe-based confocal laser endomicroscopic (pCLE) evaluation in the biliary system.

By applying all of these techniques, we were able to detect an obstruction of mucoid material in the biliary tree. The brushings and biopsies taken in the left ducts were positive for high grade intraepithelial neoplasia.

Magnetic resonance cholangiopancreatography (MRCP) demonstrated that both the intra- and extra-hepatic biliary ducts were dilated (max 22mm in the common bile duct), thus suggesting Biliary Tract (BT)-IPMN with abundant intraluminal mucous secretions. Concomitant elevations of Ca 19.9 (226U/ml), AST/ALT (128/91U/L), gGT (454U/L) and bilirubin tot/dir (10.2/9.9mg/dL) were observed.

In order to determine whether the neoplasia also extended to the right hepatic ducts we decided to perform another ERCP with Spy Glass DS System; this would provide essential information in order to consider a possible surgical approach with left hepatectomy.

At first, the ERCP highlighted a massively dilated CBD as well as minimal visibility of the left hepatic ducts after contrast injection. A 2cm filling defect was also seen at the helium. Cholangioscopy using the Spy Glass DS System showed a large amount of mucus all the way up the CBD to the biliary bifurcation, where a whitish occluding-like mass was seen. Several pCLE scans were performed via a cholangioflex probe and did not show any sign of papillae nor other neoplastic abnormalities. Targeted biopsies were made in the right ducts as well as at the bifurcation, since the whitish hilar mass was no longer detectable. The mucosa showed only endoscopic signs of chronic inflammation.

Histologic examination showed a chronic inflammatory cell infiltration of the biliary mucosa, with no dysplasia.

After some months, the patient underwent a hepatic resection of left liver lobe. Histologic examination of the surgical specimen demonstrated a multinodular, partly cystic mucinous mass. It was determined that the mass was BT-IPMN with intermediate and severe intestinal dysplasia, combined with an invasive part (5% of the whole mass) defined as moderately differentiated carcinoma (G2), with mucinous aspects (colloid carcinoma). The surgical margin was tumor free.

The patient, now with a percutaneous biliary drainage, has not experienced any more episodes of cholangitis or elevation of cholestasis indexes. As for today, there is no evidence of relapse of the disease.

Conclusion

Cholangioscopy together with other ancillary techniques enabled a definitive diagnosis of BT-IPMN in the left hepatic system. They also mostly excluded the suspect of IPMN and malignancy in the right intra hepatic biliary tract. This diagnosis enabled a surgical approach, which fortunately was radical, confirming the data obtained with multiple intraductal techniques approach in such a challenging diagnostic case.
