

Papillomatosis of Bile Duct: Case Report

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Abstract

Case Report

The papillomatosis of bile ducts is a rare disease, we report a case of a 60 year old man treated in our unity for this pathology to illustrate diagnostic difficulty and the particularities of this pathology and to show the interest of chirurgicall resection as a traitement before malignant transformation.

Keywords: Papillomatosis Bile Duct.

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INTRODUCTION

The papillomatosis of bile ducts is a rare disease, it's a precancerous condition. The diagnosis is often difficult; we are reporting a case in our unit illustrating this diagnostic difficulty and the peculiarities of this pathology.

CASE REPORT

A 60 year old ASA II man followed for type II diabetes and for glaucoma, the initial symptomatology was made of two months of evolution of a pain in the right hypochondrium associated with a fever and chills, then brutal appearance of an mucocutaneous frank

jaundice with dark urine and discolored stool and pruritus, this symptomatology regressed after 24 hours, the general state was preserved.

The patient was taken care of by an gastroenterologist having performed an abdominal ultrasound who objectified a litiasis vesicular with a thin-walled gallbladder, The MRI has objectified a 13mm litiasis vesicular, a 12mm dilated main bile duct with the presence of two evocative images of microlithiasis with a low bile duct measuring 3.55 and 44 mm respectively associated with balthazard pancreatitis stage B.



MRI findings

In this moment the diagnosis of lithiasic pathology was posed with indication of a first endoscopic sphincterotomy, then laparoscopic cholecystectomy, The ERCP found a lacunar image of a low bile duct of 9mm, sphincterotomy then extraction

of tissue material which anatomopathological analysis revealed a intraductal papillary neoplasia of the bile ducts gastro-biliary type with high grade dysplasia lesions.

After a multidisciplinary concertation meeting, an indication for surgical treatment was made. On admission the patient was not jaundiced but still had pain. The biological assessment highlighted a total bilirubin at 15, GGT at 43, ALP at 81, HB at 13, GB at 5000; the operability assessment noted no contraindication to surgery, the patient benefiting from a cephalic duodeno-pancreatectomy. The after operations noted an wound infection treated by a parietal drain and antibiotic therapy and having regressed on day + 12.

The anatomopathological examination of the operating part piece a dual location of an intraductal biliary neoplasia Of 2 and 1cm with a high grade dysplasia. The limits are passed in healthy zone and absence of metastatic nodes. Also absence of invasive elements of vascular emboli and perineural sheathing. The chemotherapy was not indicated with this patient.



Operating piece with opening of the main bile duct: presence of two polypoid lesions of low common bile duct

DISCUSSION

The papillomatosis of bile ducts or intraductal papillary neoplasm of the bile duct (IPNB) are rare diseases of the biliary tract characterised by the distinctive papillary proliferation of the bile duct epithelial cells around the slender fibrovascular stalks [1]. The diagnosis is often difficult is revealed by the anatomopathological report of the operating piece, it was first described by Biliary papillomatosis was described in 1894 by Chappet [2], and in 1959, Caroli [3] gave an anatomic description. It is most frequently observed in middle-aged and elderly patients, possessing a male-to-female ratio of 2:1 [4] and the main concern is the malignant transformation of the tumor into adenocarcinoma, which occurs in 83% of cases [5].

The symptomatology is often difficult made of an intermittent cholestatic jaundice which can be complicated by cholangitis, a table simulated that of lithiasis of the main bile duct, this can be explained by the friable consistency of the adenoma [5,6].

pre-operative diagnosis is difficult: the computed tomography or ultrasound suggest the diagnosis of papillomatosis of bile ducts in typical forms, showing an enlarged biliary tree with an irregular wall, lined with polypoid formations. The retrograde cholangiography by endoscopic way visualizes an irregularity of the biliary surface with non-mobile gaps, and allows biopsies (or biliary brushing) and the placement of a stent [7].

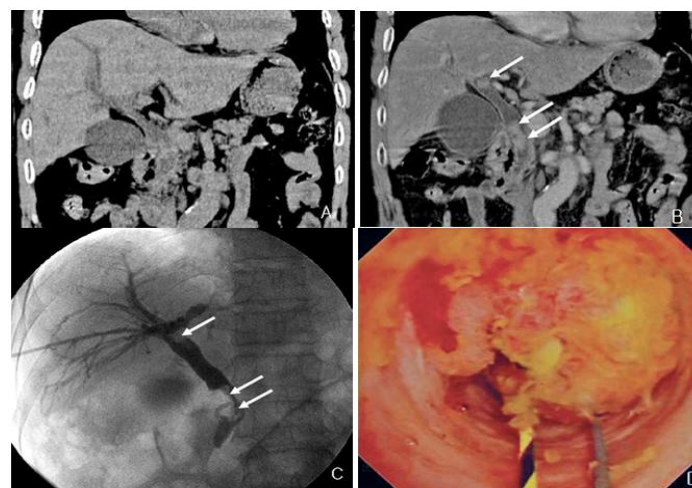


Fig-2: A 74-year-old man with epigastric pain for 1 month. A) Reconstructed coronal precontrast and B) postcontrast CT images showing a dilated extrahepatic bile duct with three abnormal intraductal papillary protruding masses in the hilar portion at the level of the common bile duct (arrows). C) PTBD tubography showing the dilatation of extrahepatic bile ducts with multifocal filling defects in the hilar portion

and common bile duct (arrows). D) PTCS shows intraluminal cauliflower-like papillary masses. e biopsy results indicated a well-differentiated papillary adenocarcinoma [7].

The risk of malignant transformation of biliary papillomatosis is 20%–50% [8,9] and can up to 83%. The progression from benign to malignant disease may follow the adenoma–carcinoma sequence.

Radical excision is typically the recommended treatment. Curative surgical resection has been observed to result in a 5 year survival rate of up to 81% [10]. In our case a cephalic duodenopancreatectomy was performed.

When surgery was not an option, local ablation with photodynamic therapy or laser via an endoscopic procedure was reported for the palliative treatment of malignant neoplasms of the bile duct [11]. Intraluminal brachytherapy with Iridium-192 has also been applied.

Anatomopathological findings [12]: Hiroaki SMD and al, examined 30 MPBTs and classified them into two distinct morphologic categories: 22 cases of "columnar type" composed of pseudostratified columnar cells with basophilic cytoplasm and columnar nuclei and 8 cases of "cuboidal type" composed of pancreaticobiliary and/or oncocyctic pattern. Pancreaticobiliary pattern showed abundantly branched papillae lined by acidophilic cuboidal cells with round nuclei, whereas oncocyctic pattern was characterized by intraepithelial lumina and cribriform pattern composed of abundant oxyphilic cells with round nuclei, and these patterns overlapped frequently. There were significant differences in the clinicopathologic findings including macroscopic findings, morphometric data, mucin expression profiles (MUC2 expression in columnar type and MUC6 expression in cuboidal type), and cell proliferative activities between columnar type and cuboidal type. Patients with columnar type showed significantly poorer survival than those with cuboidal type. We concluded that columnar type and cuboidal type of MBPTs belong to different lineage of neoplasm and that they are counterparts.

CONCLUSION

Biliary papillomatosis is a rare disorder characterised by multiple papillary adenomas in the biliary tree. It affects mainly middle-aged, or elderly persons and commonly presents with obstructive jaundice and cholangitis. The papillomatosis varies in extent and distribution within the intrahepatic and/or extrahepatic biliary tree. The papillomas can be classified into mucin or non-mucin secreting, and are premalignant with definite malignant potential. The pathogenesis of this condition is unknown, although it has been suggested that the malignant transformation follows the pathway of adenoma to carcinoma sequence, similar to colonic polyps adenoma. The definitive treatment is surgical resection.

FOOTNOTE

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this Case Report and any accompanying images.

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