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Case report

Unusual presentation of gallbladder papillomatosis without bile duct involvement: A case report ☆☆☆★★

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ABSTRACT

The papillomatosis is a very rare benign pathology diagnosed histologically with a significant potential for malignant transformation. We report a case a 60-year-old female without comorbidity present the gallbladder papillomatosis without involvement of the intra or extra hepatic biliary tract. The interest in knowing the radiological aspect of this pathology and make the early diagnosis in order to oriented treatment.

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Introduction

Gallbladder or bile duct papillomatosis is a rare disease entity characterized by multiple mucin or non-mucin secreting papillary adenoma in the biliary tract, with a significant malignant transformation potential found in 35% of cases [1–3]. Biliary papillomatosis pathogenesis continues to be elucidated; however bile stasis and recurrent infection induced by hepatolithiasis likely contribute to the chronic inflammation and subsequent mucosal changes that lead to biliary papillomatosis [3].

We report a case of papillomatosis of the gallbladder with its typical appearance on CT and MRI.

Clinical observation

An 60-year-old female presented to the emergency department for right epigastric pain with fever and vomiting for 3 days without comorbidity. The pain was nonradiating and was not related to eating or positional changes. The patient

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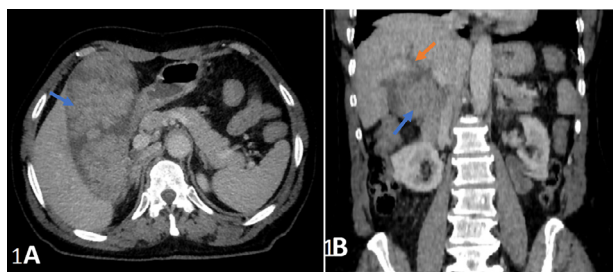


Fig. 1 – Abdominal CT scan in axial (A) and coronal (B) section with injection of contrast shows a distended gallbladder with polypoid formations of tissue density (blue arrow), associated with a slight dilation of the intrahepatic bile ducts (orange arrow). Color version of figure is available online.

was afebrile without scleral icterus. Abdominal examination found tenderness in the right hypochondrium with a positive murphy's sign. The remainder of the physical examination was unremarkable.

Laboratory data shows elevated liver enzymes, total and direct bilirubin.

Abdominal ultrasound shows a distended gallbladder with endoluminal tissue formations of intermediate echogenicity without a posterior shadow cone, associated with slight dilation of the intrahepatic bile ducts.

The abdominal CT scan with contrast injection reveals a thin-walled distended gallbladder, containing polypoid formations of tissue density, associated with a slight dilation of the intrahepatic bile ducts (Fig. 1A and B).

On MRI, the lesions appear in hypointense on T1-weighted (Fig. 2A), intermediate signal on the T2-weighted (Fig. 2B) and hyperintense on diffusion-weighted (Fig. 2C). The lesions do not enhance significantly after administration of Gadolinium (Fig. 2D). Magnetic resonance cholangiopancreatography (MRCP) shows a discrete dilation of the intrahepatic bile ducts with respect for the main bile duct and the Wirsung duct (Fig. 3A and B).

The patient underwent a surgical resection with a histological study, which confirmed the diagnosis of papillomatosis of the gallbladder without malignant transformation.

Discussion

The papillomatosis is a very rare pathology diagnosed histologically with a frequency of age between 50 and 60 years. It is a diffuse hyperplasia of the mucus made up of multiple villi developed at the expense of the biliary epithelium, of variable distribution in the intra and / or extra hepatic bile ducts [1,2].

In most cases, papillomas are histologically benign, but there are forms with malignant degeneration [3]. It is a pathology with potential for malignant development with a risk of malignant transformation into cholangiocarcinoma estimated between 11 and 41% [1].

Two types of papillomatosis are described: that without secretion of mucin and that with secretion of mucin. These lat-

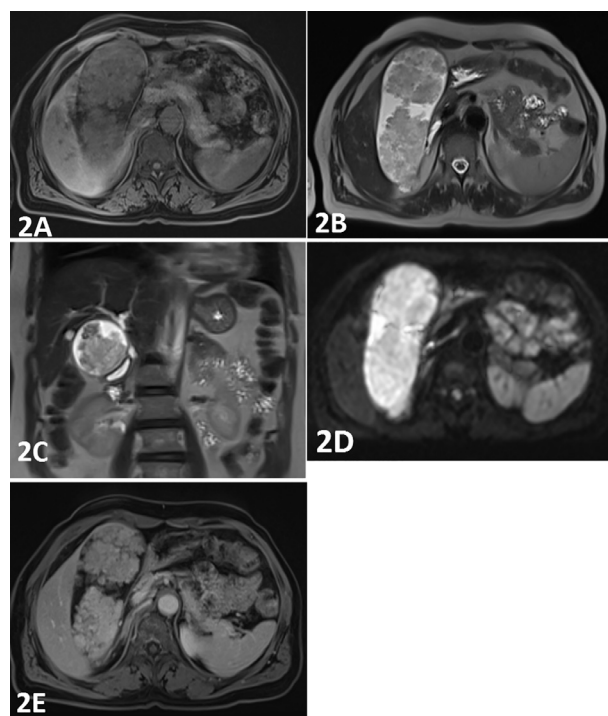


Fig. 2 – MRI demonstrate the lesions: in hypo signal in axial T1 section (A). An intermediate signal on the T2-weighted sequences in the axial (B) and coronal section (C). A restriction to diffusion image in axial section (D). The lesions do not enhance significantly after administration of Gadolinium in axial section T1 fat sat after injection of gadolinium (E).

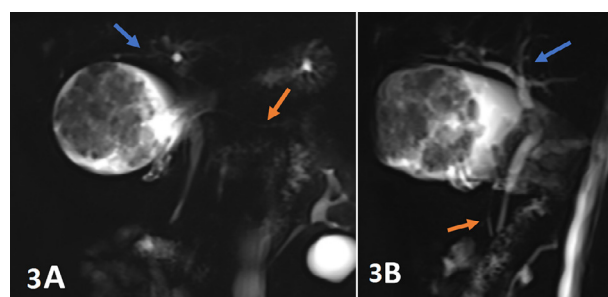


Fig. 3 – Magnetic resonance cholangiopancreatography in face (A) and oblique (B) section shows a discreet dilation of the intrahepatic bile ducts (blue arrow) with respect for the main bile duct and the Wirsung duct (orange arrow). Color version of figure is available online.

ter lesions have the ability to produce a large amount of mucus that can completely block the bile ducts and cause dilations. The secreting form has similarities to intraductal papillary and mucinous tumors of the pancreas (TIPMP) [2,3].

The clinical signs are not specific. The patient may be completely asymptomatic as he may manifest nausea, right hypochondrial pain, or even repeated episodes of cholangitis in the event of associated involvement of the bile ducts [1,4].

Imaging plays an important role in guiding the diagnosis when the image is typical. Abdominal ultrasound shows fairly limited and distinct endoluminal tissue images of intermediate echogenicity without a posterior shadow cone, which may be associated with dilation of the upstream bile ducts in cases of bile duct involvement. The abdominal CT scan with injection of contrast shows a distended gallbladder, lined with hypo dense polypoid formations sometimes associated with dilation of the bile ducts with irregular thick walls. [3–5]

Magnetic resonance imaging is the best way to analyze biliary dilation and its contents, whether or not the Wirsung canal is respected. On Bili-MRI, the lesions of papillomatosis appear as hypo intense in the T1 sequence and slightly hyperintensity on the T2-weighted sequences. The lesions do not significantly enhance after administration of Gadolinium, and remain in hypo-signal to the adjacent hepatic parenchyma. Magnetic resonance imaging allows a better analysis of biliary dilation and its content, whether or not the Wirsung canal is respected. [3–5]

Endoscopic retrograde cholangiography may show secretion of mucus through the papilla of the ampulla of Vater and images of lacunae. This test also allows cytological analysis, but its ability to assess the extent of the disease is underestimated, due to the presence of mucus, preventing the diffusion of the contrast [1].

Cholangioscopy allows a direct examination of the lesions, specifies the extent of the papillomatosis and allows the different treatments to be adapted. The interest of this examination also lies in the realization of multiple biopsies confirming the diagnosis, but does not allow to exclude the diagnosis of cholangiocarcinoma adjacent to the biopsies. On the other hand, unlike cytology, the analysis of the biopsies makes it possible to specify the degree of dysplasia and malignant transformation in case of invasion of the mucous chorion [1,4,5].

Localized papillomatosis can benefit from surgical resection, completed intraoperatively by ultrasound associated with choledocscopy. The risk of recurrence appears to be

high, partly explained by the failure to perform choledocscopy pre- and / or intraoperatively. Cephalic duodenopancreatectomy with upper biliary-enteric anastomosis is indicated when the papillomatosis extends to the distal bile duct or proximal Wirsung's duct [4,5].

Conclusion

Papillomatosis of the gallbladder or bile ducts in general is a rare disease with a significant potential for malignant transformation, especially for the mucus-secreting type. Hence the interest in knowing the radiological aspect of this pathology in order to make the diagnosis precociously and guide treatment.

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