CLINICAL IMAGE



Bifocal intracholecystic tubulopapillary neoplasm: A rare and distinct tumor of the gallbladder

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Abstract

Intracholecystic papillary-tubular neoplasms are rare precancerous and polypoid lesions of the gallbladder, found in nearly 0.4% of cholecystectomy specimens. They follow the adenoma-carcinoma sequence. About half the cases are associated with invasive carcinoma. Their diagnosis relies on histopathological examination with extensive sampling of the surgical specimen.

KEYWORDS

gallbladder cancer, intracholecystic papillary neoplasm, laparoscopic cholecystectomy

CLINICAL IMAGE

A 76-year-old man with diabetes and coronary artery disease consulted for right upper quadrant pain and fever. 1,2 Abdominal CT scan showed a slight wall thickening of the gallbladder fundus and a polypoid lesion, which enhanced moderately after injection of contrast product (Figure 1A). The patient underwent laparoscopic cholecystectomy. Macroscopically, there were two polypoid lesions near the neck and at the fundus measuring 1.3 and 1.5 in diameter, respectively (Figure 1B,C). The surrounding mucosa was granular and rough (Figure 1B,C).

Frozen section analysis of the gallbladder established the diagnosis of low-grade adenomas. Hence, liver tissue resection from segment IVb and V was not performed.

Deferred histological examination of the cholecystectomy specimen after formalin fixation established the diagnosis of intracholecystic tubulo-papillary tumor of biliary type with low-grade dysplasia (Figure 1D) for the first polyp, whereas the second polyp corresponded to an intracholecystic tubulo-papillary tumor of biliary type with high-grade dysplasia associated with an invasive carcinoma invading the muscularis (Figure 2A,B). In the surrounding mucosa, we noted high-grade biliary intraepithelial neoplasia (Figure 2C) as well as intestinal and pyloric metaplasia (Figure 2D). Extended liver tissue resection from segment IVb and V was planned for this patient who was lost to follow-up.

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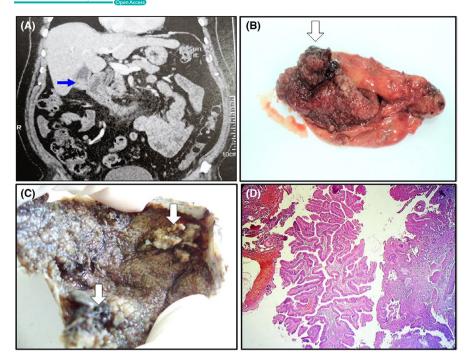


FIGURE 1 (A) Abdominal CT scan showed a slight wall thickening of the gallbladder fundus and a polypoid lesion (blue arrow). (B) Macroscopic examination of the incompletely open cholecystectomy specimen (before formalin fixation) revealing a polypoid lesion of the gallbladder fundus (white arrow) associated with granular lesions in the surrounding mucosa. (C) Macroscopic examination of the cholecystectomy specimen (after formalin fixation) revealing two polypoid lesions at the fundus and near the neck of the gallbladder measuring respectively 1.5 and 1.3 cm in diameter (white arrows) associated with granular lesions in the surrounding mucosa. (D) Intracholecystic tubulo-papillary tumor of biliary type with low-grade dysplasia (Hematoxylin and eosin, ×40)

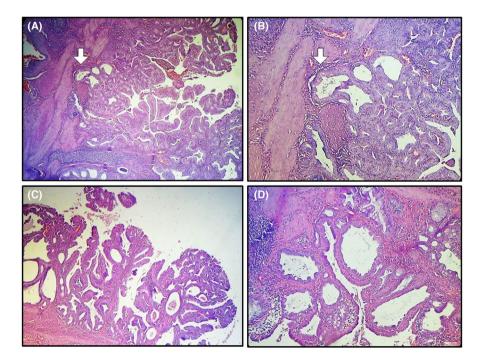


FIGURE 2 (A) Intracholecystic tubulo-papillary tumor of biliary type with high-grade dysplasia associated with an invasive carcinoma invading the muscularis (white arrow; Hematoxylin and eosin, ×40). (B) Intracholecystic tubulo-papillary tumor of biliary type with high-grade dysplasia associated with an invasive carcinoma invading the muscularis. Tubulo-papillary growth pattern with back-to-back epithelial units and limited stroma. Invasion of the muscularis was focally noted, (white arrow; Hematoxylin and eosin, ×200). (C) In the surrounding mucosa, we noted high-grade biliary intraepithelial neoplasia with a papillary pattern (Hematoxylin and eosin, ×40). (D) Intestinal and pyloric metaplasia were noted in the surrounding mucosa (Hematoxylin and eosin, ×200)

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Dr Faten Limaiem and Dr Sahir Omrani prepared, organized, wrote, and edited all aspects of the manuscript. Dr Faten Limaiem prepared all of the histology figures in the manuscript. Dr Sahir Omrani participated in the conception and design of the study, the acquisition of data, analysis and interpretation of the data. All authors contributed equally to preparing the manuscript and participated in the final approval of the manuscript before its submission.

ETHICAL APPROVAL

All procedures performed were in accordance with the ethical standards. The examination was made in accordance with the approved principles.

CONSENT

Published with written consent of the patient.

DATA AVAILABILITY STATEMENT

In accordance with the DFG Guidelines on the Handling of Research Data, we will make all data available upon request.

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REFERENCES

- 1. Shimada S, Homma T, Koyanagi K, et al. Intracholecystic papillary neoplasm of the gallbladder diagnosed during follow-up of Menetrier's disease: a case report. *Mol Clin Oncol*. 2021;15(5):233.
- 2. Kinuthia KG, Kodiatte TA, Burad D, et al. Intracholecystic papillary tubular neoplasms of the gallbladder. A clinicopathological study of 36 cases. *Ann Diagn Pathol*. 2019;40:88-93.

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