

An Unexpected Delivery

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Received Date: October 16, 2023 | Accepted Date: November 24, 2023 | Published Date: December 01, 2023

Citation: Wishahi E., Mohamad A. Arisha, Fisher Y., Klein A., (2023), An Unexpected Delivery, *International Journal of Clinical Case Reports and Reviews*, 15(3); DOI:10.31579/2690-4861/342

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Abstract:

Background: Intraductal Papillary Mucinous Neoplasms of the Bile Duct (IPNBs) are rare biliary tumors. We present an unusual case with its management.

Case Presentation: A 58-year-old man with a history of metastatic TCC presented with recurrent biliary abdominal pain and pruritus. Imaging revealed a dilated common biliary duct with a stone-like filling defect. ERCP revealed a grape-like polycystic mass, subsequently diagnosed as IPNB.

Conclusion: IPNBs, though rare, should be considered in the differential for atypical biliary lesions. Timely diagnosis and management are essential due to their malignant potential.

Key words: IPNB (intraductal papillary mucinous neoplasm of the bile duct); ERCP (endoscopic retrograde cholangiopancreatography); biliary stone; high-grade dysplasia; biliary tract tumors

Introduction

Intraductal Papillary Mucinous Neoplasms of the Bile Duct (IPNBs) are a rare cause of biliary obstruction. These tumors, though infrequent, can present in various manners, sometimes making diagnosis and management challenging.

Materials and Methods:

A 58-year-old man with a history of metastatic TCC presented with recurrent biliary abdominal pain and pruritus. The patient underwent a series of diagnostic evaluations, including blood tests, Abdominal Ultrasound, and MRCP. The MRCP showed a filling defect in the distal CBD suspected to be a stone. ERCP, was performed for both diagnostic and therapeutic purposes.

Case Presentation

A 58-year-old man, presented to the hospital with recurring biliary abdominal pain and pruritus, which began 3 months prior to his admission. He had no jaundice or fever, no vomiting and no weight loss. Four years previously, he was diagnosed with metastatic TCC to kidney and liver. He underwent chemoradiation therapy and a recent PET-CT showed no evidence of active disease.

Physical examination was normal. Laboratory data revealed high cholestatic liver enzymes with a normal bilirubin level. Abdominal Ultrasound, showed a dilated common biliary duct (CBD) up to 13 mm. MRCP also showed a dilated CBD with a filling defect in the distal CBD consistent with a stone (Figure1).

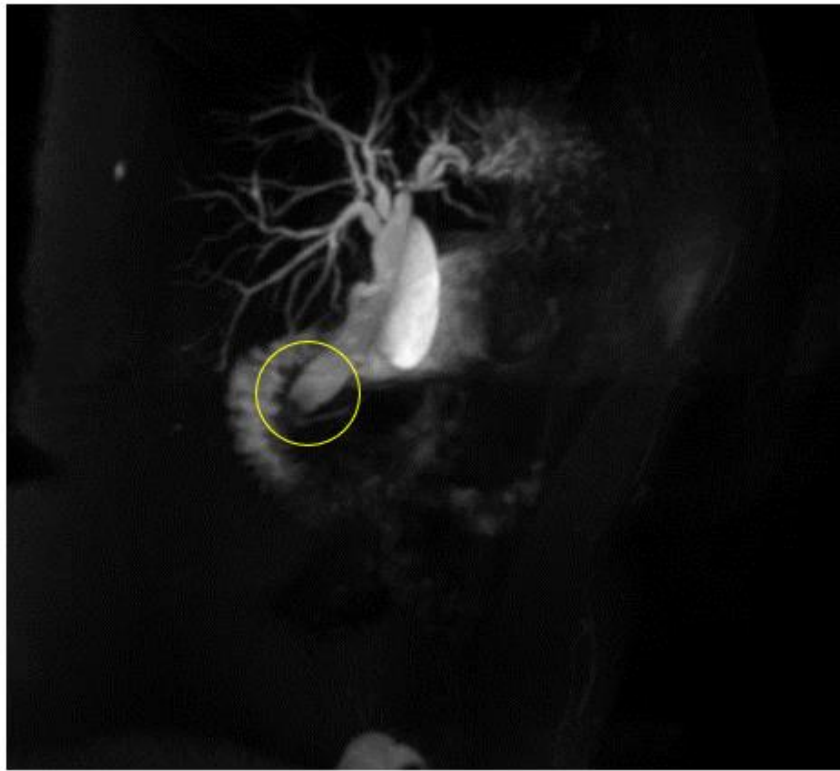


Figure 1: Dilated CBD up to 13 mm and a distal CBD filling defect.

The patient was referred to ERCP. The papilla appeared bulky with an “impacted stone” appearance. CBD cannulation was performed and fluoroscopy demonstrated a dilated CBD up to 14 mm with a distal filling defect (Figure 2). After papillotomy, a stone extraction balloon was

passed and a large grape-like polycystic mass was delivered from the bile duct (figure 3). A prophylactic stent was placed and the mass was resected en-bloc with a hot snare and retrieved for pathology.

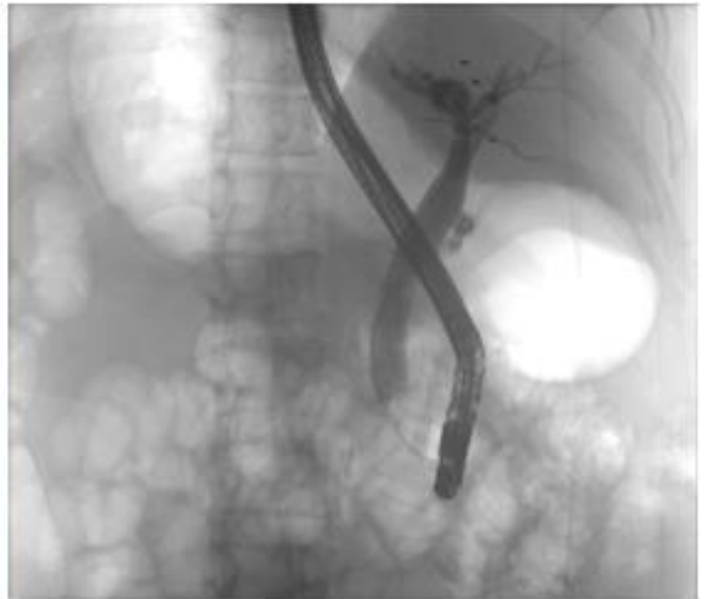


Figure 2: Endoscopic retrograde cholangiography showing a bulky papilla with normal overlying mucosa. The CBD is dilated up to 14 mm with a distal amorphous filling defect.

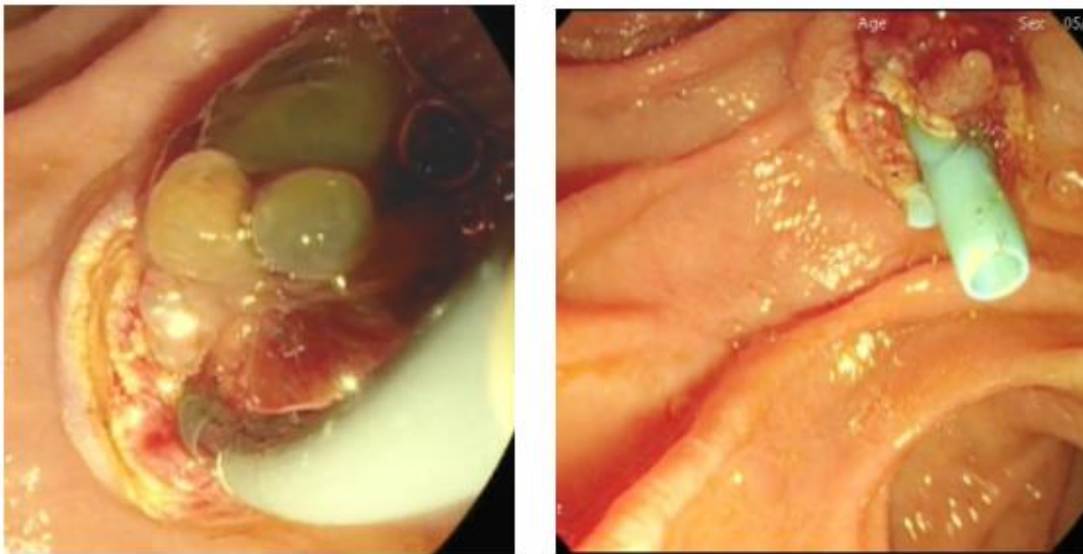


Figure 3: After papillotomy, a balloon has been transferred and a large grape-like polycystic mass has emerged. Stent was placed.

The differential diagnosis included: Echinococcal disease (patient had no relevant epidemiological history); Metastatic RCC (as mentioned, PET-CT recently was clear); Intraductal papillary mucinous neoplasms of the

bile duct (IPNBs). The final diagnosis was IPNBs defined histologically by glandular tumor with mucus secretion and areas of high-grade dysplasia (Figure 4).

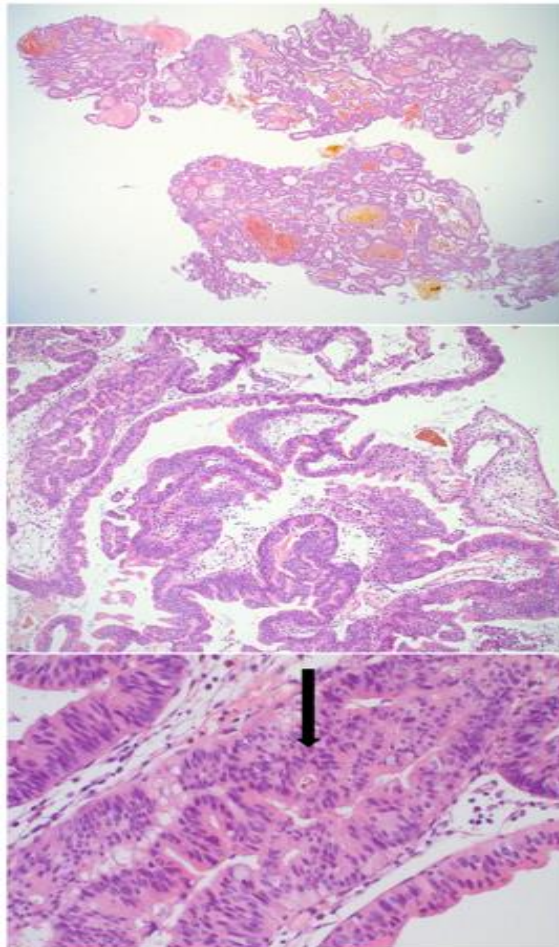


Figure 4: A [x12.5] Biliary IPMN showing papillary structures on the surface epithelium and complex glandular structures. B [x100] Focal structures show a complex architecture with nuclear pseudostratification. C [x400] Foci of high-grade dysplasia with nuclear atypia and loss of polarity. Arrow shows focal necrosis.

IPNB is a rare disease, and its invasive form, papillary cholangiocarcinoma, accounts for approximately 4%-38% of all bile duct adenocarcinomas. IPNB most commonly develops in patients between 50 and 70 years of age. The pathogenesis and progression pattern have not yet been well defined, but its clinic-pathologic features are similar to IPMN of the Pancreas. Curative resection is the preferred primary treatment.

Discussion:

Intraductal Papillary Mucinous Neoplasms of the Bile Duct (IPNBs) represent a rare subset of biliary tumors with varied clinical presentations, making their diagnosis and management challenging [1-2].

This case underlines the diagnostic complexities associated with IPNBs [4].

The unique aspect of this case was the unexpected delivery of the polycystic tumor from the CBD during ERCP mimicking a conventional biliary stone [5].

The endoscopic appearance during the ERCP was considered other possibilities, such as Echinococcal disease, or metastatic Renal Cell Carcinoma (RCC). The lesion was completely resected with hot snare and the final histology showed IPNBs with glandular tumor features and high-grade dysplasia provided a definitive diagnosis [7].

IPNBs, although rare, share clinical and pathological characteristics with Intraductal Papillary Mucinous Neoplasms of the Pancreas (IPMN) [6]. This case highlights the importance of considering IPNBs in the differential diagnosis of atypical biliary obstructions and the need for early diagnosis due to their malignant potential [7].

Conclusion:

In conclusion, this case demonstrates the diagnostic complexities of Intraductal Papillary Mucinous Neoplasms of the Bile Duct (IPNBs). Early diagnosis and a multidisciplinary approach are crucial given the potential for malignancy.

Funding:

None.

Competing interests:

None.

Patient and public involvement: None.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

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DOI:10.31579/2690-4861/342

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