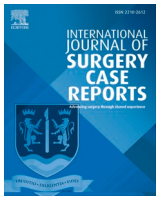
International Journal of Surgery Case Reports 115 (2024) 109243

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report

Intraductal papillary neoplasm of the bile duct: A case report of a rare tumor with a brief review of literature

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ARTICLE INFO

Keywords:
Intraductal papillary neoplasm
Invasive component
Hepatectomy
Pancreato-biliary type

ABSTRACT

Introduction and importance: Intraductal papillary neoplasm of the bile duct (IPNB) is a rare neoplasm, mostly found in patients from far Eastern areas where hepatolithiasis and clonorchiasis are endemic. Very few cases are reported from India.

Case presentation: We present a case of recurrent cholangitis in a 59-year-old male, initially suspected to have IPNB based on preoperative imaging. Confirmation occurred intraoperatively, with mucin-filled bile ducts and papillary lesions in the resected hepatic duct. Treatment included left hepatectomy, extrahepatic bile duct excision, and Roux-en-Y hepaticojejunostomy. Histopathology indicated invasive pancreatobiliary-type IPNB with clear margins. The patient experienced post-hepatectomy hepatic insufficiency and superficial incisional surgical site wound infection, managed conservatively. Discharge occurred on postoperative day 21, with satisfactory recovery at the 16-month follow-up.

Clinical discussion: IPNB is recognized as the biliary equivalent of intraductal papillary mucinous neoplasm, as these two conditions exhibit multiple commonalities in terms of clinical and histopathological characteristics. The unique aspect of our case lies in the intricacies associated with its diagnosis. Initially, imaging modalities did not yield a definitive characterization of the lesion. Notably, the endoscopist misinterpreted mucin expression emanating from the papilla as purulent material, primarily due to the patient's concurrent cholangitis. Subsequent repetitions of both CT scan and MRI provided some valuable insights that contributed to the diagnostic clarity of the IPNB.

Conclusion: In cases of symptoms like biliary obstruction with bile duct dilation, wall nodules, papillary/solid-cystic masses, and upstream-downstream dilation, IPNB should be considered. Striving for R0 resection is crucial for enhanced long-term patient survival.

1. Introduction

Intraductal papillary neoplasm of the bile duct (IPNB) is a rare neoplasm of the bile duct characterized by intraductal papillary or villous neoplasms covered by neoplastic epithelium with fine fibrovascular stalks in the dilated bile duct [1,2]. It is mostly found in patients from far Eastern areas where hepatolithiasis and clonorchiasis are endemic [3,4]. IPNB may occur anywhere in the biliary tree. Most commonly it involves the left lobe of the liver [5,6]. Common presentations are right upper abdominal pain, jaundice, and acute cholangitis [5]. Most patients are between 50 and 70 years of age with a slight male predominance [6,7]. It has high malignant potential with invasive components being found in 30 to 80 % of patients [5,6,8–10]. Depending upon the cellular pattern, four subtypes have been described:

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https://doi.org/10.1016/j.ijscr.2024.109243
Received 27 October 2023; Received in revised form 3 January 2024; Accepted 5 January 2024
Available online 15 January 2024

gastric, intestinal, pancreato-biliary, and oncocytic. Pancreato-biliary is the most common subtype with the highest rate of invasive components [5,6,8,9]. IPNB is regarded as the biliary counterpart of the intraductal papillary neoplasm of the pancreas (IPMN) [10]. Surgery is considered the cornerstone of treatment. Due to the high risk of malignant transformation and high rate of loco-regional recurrence, R0 resection is recommended for better long-term survival [11]. Overall prognosis of IPNB is better than conventional cholangiocarcinoma [12–14]. Herein, we report a case of IPNB with an invasive component who presented with recurrent cholangitis over a period of 3 years. The work has been reported in line with the SCARE criteria [15].

2. Case presentation

A 59-year-old male, without any medical co-morbidity, presented to our institute with a history of recurrent episodes of cholangitis for the past 3 years. He was admitted to a local hospital 3 years back with jaundice, mild upper abdominal pain, and fever with chills and rigor for 7 days. He was managed with intravenous antibiotics and an endoscopic retrograde cholangiopancreaticogram (ERCP) with biliary stenting. Subsequently, his symptoms resolved. ERCP findings at that time were dilated bile duct (14 mm) with sludge in situ. Magnetic resonance cholangiopancreatography (MRCP) findings were suggestive of a type IV choledochal cyst. Seven months later, he developed recurrence of symptoms and was re-admitted. Another ERCP and biliary stenting was performed. At this time ERCP findings were dilated extrahepatic biliary tree and left ductal system. During stent placement, pus was seen coming out from the papilla. The provisional diagnosis was recurrent pyogenic cholangitis (RPC) involving the left lobe of the liver. He was referred to our department for definitive therapy. Because of the COVID-19 pandemic, he did not come to our hospital then. He presented to our outpatient department 2 years later with severe acute cholangitis. Physical examination revealed icterus, hepatomegaly, and fever of 103 °F. Laboratory investigations revealed total bilirubin (TB) of 15 mg/dL (normal range: 0.4–1 mg/dL), alkaline phosphatase (ALP) of 1098 U/L (normal range: 60–120 U/L), AST of 78 U/L (normal range:30–40 U/L), ALT of 106 (normal range: 30–40 U/L), INR of 1.4, and total leukocyte count of 16,500/cu.mm (4000–10,000/cu.mm). Serological tests for hepatitis B and C viruses were negative. We thought that the acute cholangitis was related to stent dysfunction. He was managed with intravenous antibiotics and repeat ERCP biliary stenting. During ERCP, the previous biliary stent was not found. The endoscopist again noticed an expression of pus from the papilla during stenting. After control of the acute episode of cholangitis, the patient was investigated with repeat MRCP and triphasic computed tomography scan (CT scan) of the abdomen. MRCP revealed a dilated left hepatic duct, extrahepatic bile duct with a 23x58x36 mm cystic lesion overlapping the left hepatic duct, crowding of the peripheral ducts, left lobar atrophy, and few solid mural nodules arising from the wall of the cystic lesion (Fig. 1a, b). A triphasic CT scan of the abdomen revealed dilatation of both extrahepatic biliary tree and the left ductal system. The left hepatic duct was hugely dilated like cistern formation with few enhanced mural nodules in late-arterial phase (Fig. 2a & b). It raised the suspicion of an IPNB. Six weeks later, we planned for left hepatectomy with or without excision of the extrahepatic bile duct and Roux-en-Y hepaticojejunostomy. The day before the operation, liver function tests (LFTs) were: TB of 3.8 mg/dL, ALP of 577 U/L, AST of 86 U/L, ALT of 66 U/L, and INR of 1.2. Preoperative hemoglobin was 10.5 g/dL (normal range: 12–15 g/dL) and platelet count was 3.3 lakhs/cu.mm (normal range: 1.5–4.5 lakhs/cu.mm). At exploration, we found that the liver was cholestatic with micronodular cirrhosis. The bile duct was dilated about 1.5 cm and a biliary cystic lesion was found on the undersurface of the left lobe of the liver. As the patient had multiple biliary interventions, we tried to aspirate bile from CBD for culture sensitivity. But to our utter surprise, we failed to aspirate any amount of bile after repeated puncture. We opened the CBD and found that the entire biliary tree was filled with mucin (Fig. 3). We then thought that the cystic lesion in the left lobe was actually a mucin-secreting neoplasm. We performed a left hepatectomy, excision of the extrahepatic bile duct, and saline irrigation of the entire right ductal system till the free flow of bile was noticed. The resected specimen was sent for frozen section and it confirmed R0 resection. Then, Roux en-Y hepaticojejunostomy was performed. The resected specimen of the liver showed a dilated left hepatic duct with multiple papillary lesions within it (Fig. 4). The histological findings were consistent with intraductal papillary neoplasm of the bile duct with invasive components (Fig. 5a-f). Sections from tumor showed papillary structures with fine fibrovascular cores covered by pancreato-biliary type of epithelial cells. The cells showed mild pleomorphism (Fig. 5d & e). Fibrous core had mild chronic inflammation. Focal area of invasion was present infiltrating the surrounding stroma (Fig. 5f). In the postoperative period, the patient developed grade A post-hepatectomy hepatic insufficiency and mild wound infection (superficial incisional surgical site infection) which were treated conservatively. The patient was discharged on postoperative day 21. The patient was well at 16-month follow-up.

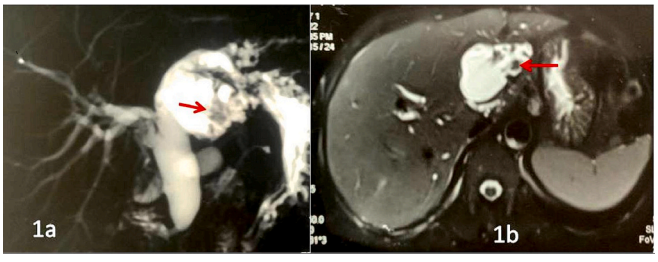

Fig. 1. a- Magnetic resonance cholangiopancreatography showed cystic dilataion of the left hepatic duct with intracystic solid component (red arrow). The extrahepatic bile duct is also dilated. b- T2 weighted image of MRI showed cystic dilatation of left hepatic duct with intracystic solid component (red arrow). Left lobe of liver is atrophied. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

3. Discussion

Intraductal papillary neoplasm of bile duct (IPNB) is a rare tumor. Initially, it was described as biliary papillomatosis and first report was published by Chappet in 1894 [16]. Multicentric biliary papillomatosis associated with invasive adenocarcinoma was first reported by Neumann et al. in 1976 [17]. Thereafter various names were used to describe this tumor like biliary papillomatosis, mucin-producing cholangiocarcinoma, mucin hypersecreting bile duct tumor, and biliary intraductal papillary mucinous neoplasm [3,8]. But, the term IPNB was coined by Zen et al. in 2006 [18]. In 2010, IPNB was included in the World Health Organization classification of bile duct tumors [19]. As there is no specific clinical feature or image finding, preoperative diagnosis is often not possible. Most patients are between 50 and 70 years of age with a slight male predominance [6,7]. Usual presentation is right upper abdominal pain and obstructive jaundice. Repeated acute cholangitis is a presenting symptom in about 5 to 59 % of patients [5]. Similarly, in our patient, the presenting feature was repeated episodes of cholangitis over a period of 3 years. Associated secondary biliary cirrhosis (SBC) in our patient can be explained by long-standing incomplete biliary obstruction with recurrent cholangitis which are considered to be the risk factors for SBC [20,21]. Up to 5 % of patients can be diagnosed in an asymptomatic state [9]. Most of the symptoms of IPNB are related to mucin producing activity as well as friable nature of the lesion. Friable tumor component can easily detach from their origin, leading to acute obstruction of the bile duct. Similarly, abundant mucin discharge from the tumor may intermittently obstruct the bile flow, leading to obstructive jaundice and cholangitis. Macroscopically, abundant mucin hypersecretion is found in only one third of patients, as

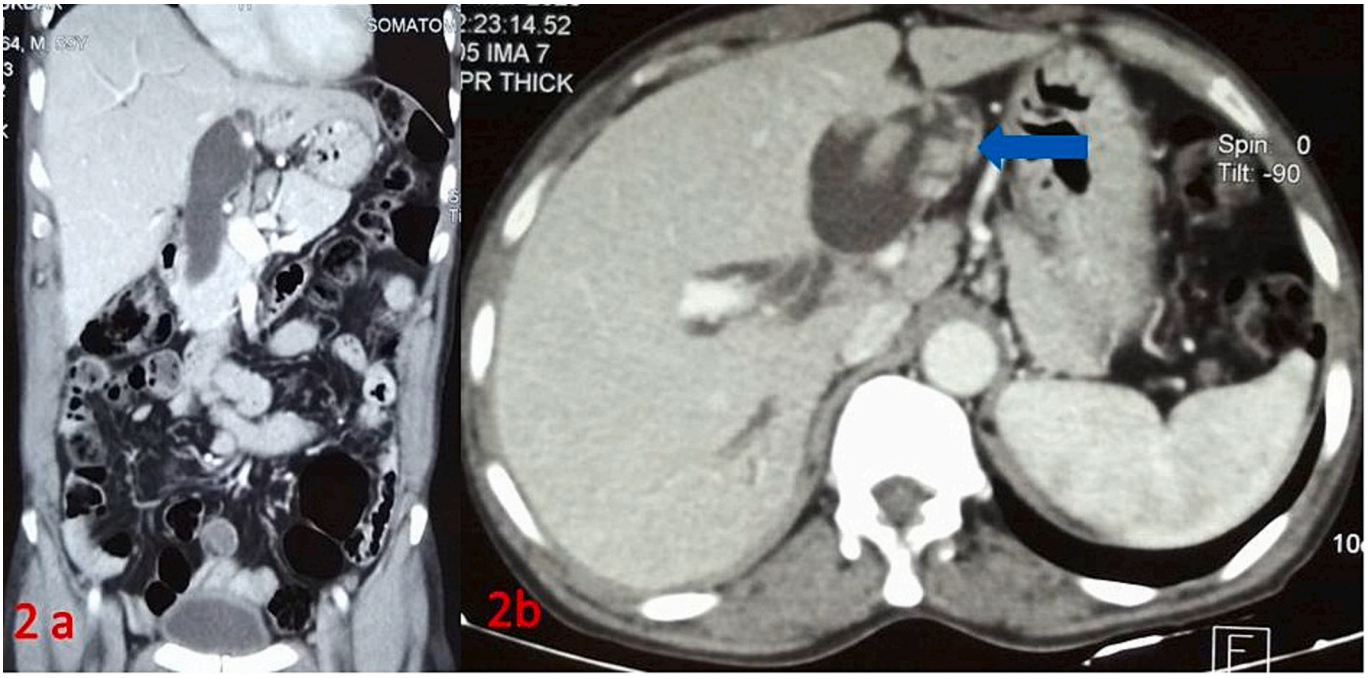

Fig. 2. a- Computed tomography scan of the abdomen (coronal view) showed dilatation of extrahepatic biliary tree as well as the left hepatic ductal system. b- cross sectional image of CT scan showed cystic dilatation of the left hepatic duct with intracystic papillary lesions enhanced in delayed arterial phase (blue arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

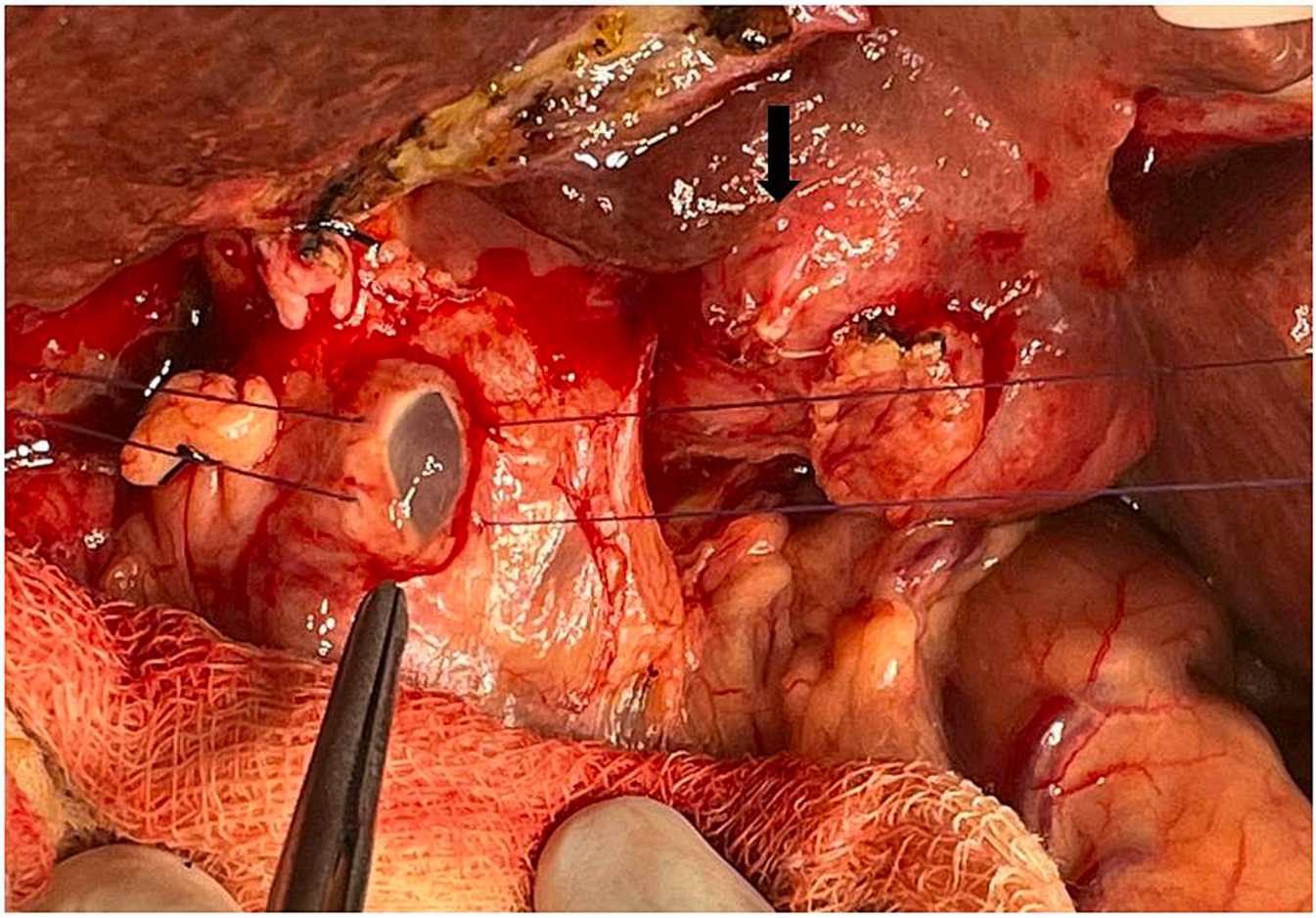

Fig. 3. Intraoperative image showed mucin filled bile duct with cystic lesion on the undersurface of the left lobe of liver (black arrow).

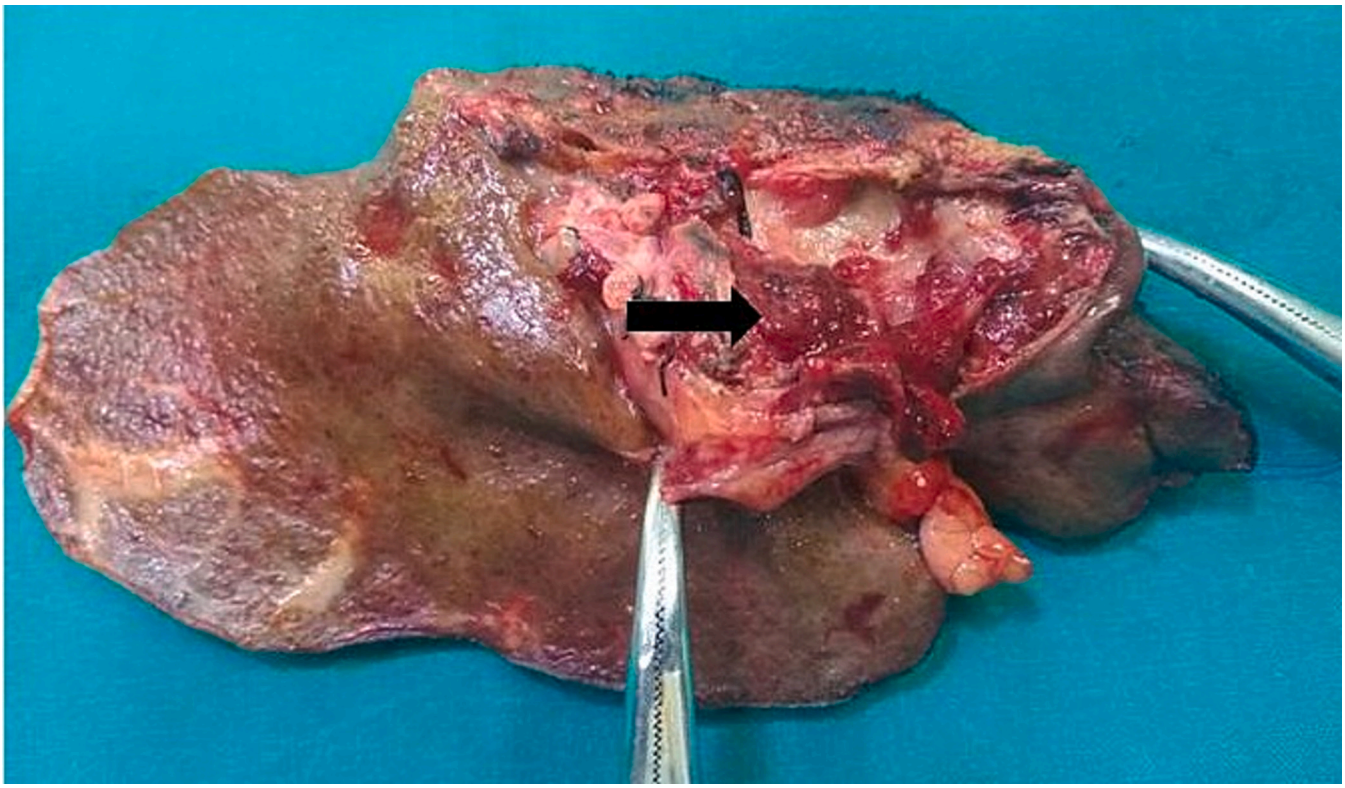

Fig. 4. Photograph of the resected specimen showed dilated left hepatic duct with intraductal papillary lesions (black arrow). The liver parenchyma showed fine nodules suggestive of secondary biliary cirrhosis.

we described in our case [11]. Another explanation of jaundice and cholangitis is associated hepatolithiasis which is regarded as an etiological factor for IPNB [3]. Yeh et al. [22] reported that nearly 87 % of patients with IPNB had hepatolithiasis in Taiwan. Kim et al. [4] from korea showed that 31 % of their patients had associated hepatolithiasis and 18 % had clonorchiasis. But, this association is rarely found in Western population [9]. The tumor can arise anywhere along the biliary tree. Nevertheless, the most frequent location seems to be the left side of the biliary tree. Tan et al. [5] analyzed 354 patients of IPNB and found that the tumors were in the left lobe in 57.5 % of patients.

The common radiologic findings for IPNB are bile duct dilatation, intraductal mass and cystic lesion [23]. Both upstream and downstream dilatation of bile duct is common in mucin overproduction type of IPNB. On triphasic CT scan papillary nodules are enhanced during late arterial phase but not in delayed phase. In MRI, it is described as hypointense mass on T1 and hyperintense mass on T2 images [7]. ERCP and percutaneous transhepatic cholangiography (PTC) can be suboptimal because excessive mucin production may prevent opacification of the entire biliary tract. Moreover, small papillomas may be missed by conventional imaging [24]. Here lies the importance of intraductal ultrasonography and cholangioscopy. Cholangioscopy allows us to directly observe the superficial spread of the lesion and detection of small lesion also [25]. Common differential diagnoses for IPNB are: mass forming intrahepatic cholangiocarcinoma, recurrent pyogenic cholangitis (RPC) with hepatolithiasis, biliary mucinous cystic neoplasm (MCN), and choledochal cyst [3,8,26]. Mass forming cholangiocarcinoma often appears as single intrahepatic mass with upstream bile duct dilatation and progressive enhancement in delayed phase of CT and MRI imaging. IPNB usually appears as multifocal papillary lesions with both upstream and downstream bile duct dilatation with or without mucin overproduction. As IPNB is suspended on fine fibrovascular stalks with scarce fibrous stroma, it rarely shows enhancement in delayed phase. MCN almost exclusively occurs in women. It rarely shows communication with bile duct. Thus, adjacent bile duct dilatation is less commonly encountered in MCN than IPNB. MCN may cause upstream biliary dilatation by direct compression but dilatation of downstream bile duct is absent. Ovarian-like stroma is the characteristic microscopic finding of MCN. It is absent in IPNB. In type IVA choledochal cyst, dilatation of both intrahepatic and extrahepatic bile ducts may occur. The major differences between choledochal cyst and IPNB are: choledochal cyst is more common in female and most of the patients become symptomatic within first decade of life. On the other hand, IPNB is more common in male and most of the patients are between 50 and 70 years of age. Choledochal cyst may be accompanied by anomalous pancreatobiliary junction (APBJ) in a significant proportion of patients. The incidence of APBJ in IPNB is similar to that of general population. If malignant transformation occurs in choledochal cyst, it is usually nodular pattern rather than papillary lesion in IPNB. RPC with stone causes intermittent biliary obstruction, biliary dilatation with filling defects on images, similarly those observed in IPNB. Mucin plugs or sloughed tumor mass may be confused with stones in conventional imaging. Therefore, it may be difficult to differentiate RPC from IPNB based on imaging alone. Sometimes, invasive methods like ERCP or cholangioscopy may be necessary to prove the presence of mucin plugs or tumor.

IPNB is regarded as a biliary counterpart of IPMN because these two lesions share several clinical and histopathologic features [8,10]. However, several important differences can be observed: first, the incidence of invasive component is more in IPNB (30–80 %) compared to IPMN (20–30 % in main duct type and 3–5 % in branch duct type); second, macroscopic mucous production is found in almost all cases of IPMN but in only one third cases of IPNB [11]; third, known risk factors (hepatolithiasis, Clonorchiasis) are present for IPNB but IPMN occurs without any obvious risk factor; fourth, pancreato-biliary subtype is the most common variety in IPNB but intestinal subtype is most common in IPMN [8].

The standard treatment for IPNB, without metastasis is radical surgical resection with adequate margins. Depending upon the location of the tumor, patient may need a major hepatectomy, excision of

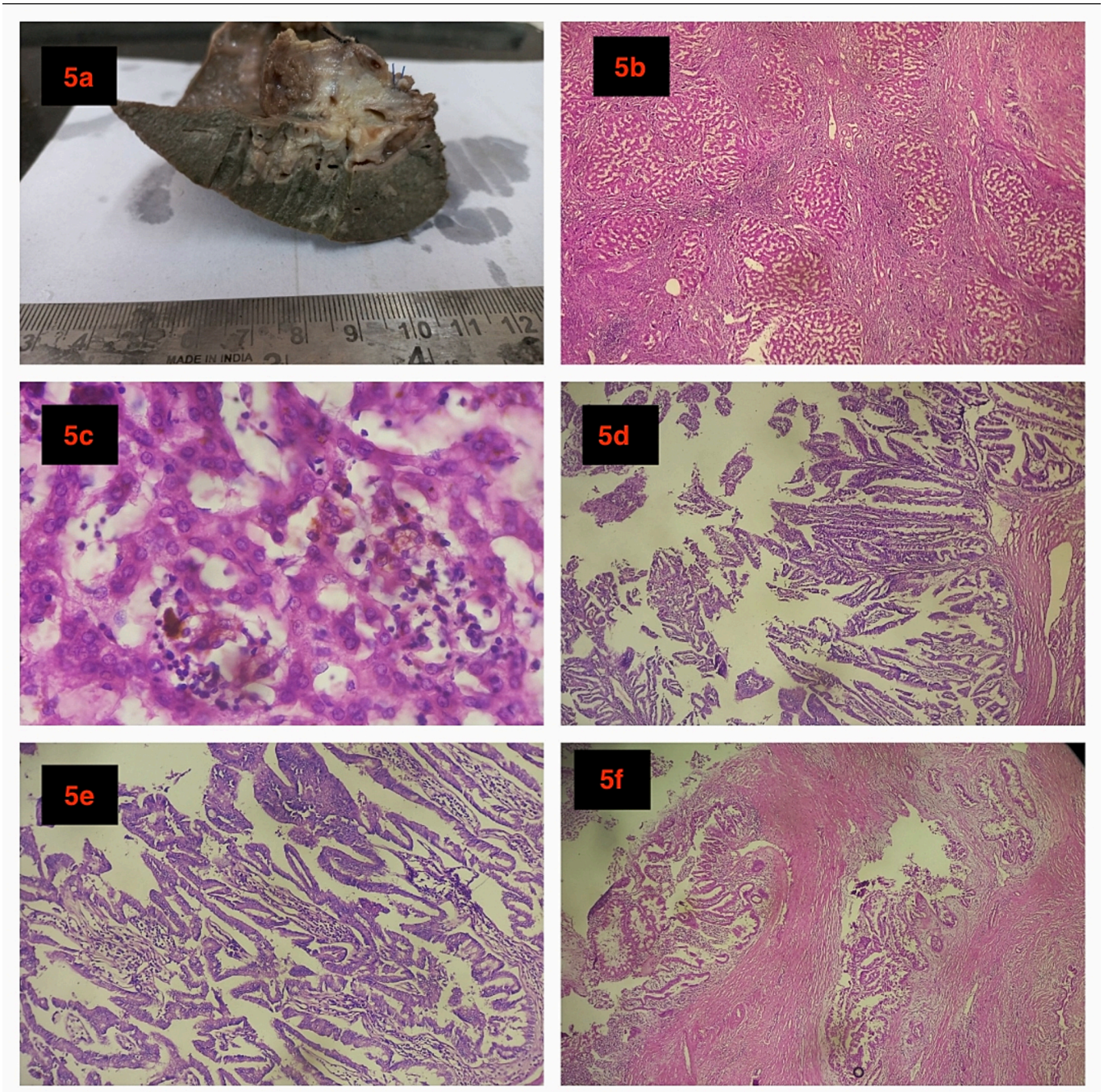

Fig. 5. Histopathology of resected specimen showed: a- Gray white fleshy tumor with irregular surface and infiltratative margins; b-Distortion of lobular architecture of adjacent liver with bridging fibrosis and irregular shaped nodule formation (HE 100×); c-Liver showing canalicular and cytoplasmic cholestasis with sinusoidal dilatation (HE 400×); d & e- Tumors with cells arranged in papillary architecture with fine fibrovascular cores covered by pancreato-biliary type of epithelial cells. The tumor cells showed nuclear stratification, nuclear hyperchromasia, and mild pleomorphism (HE 100×); f- Foci of tumor invasion with surrounding desmoplastic reaction (HE 100×).

extrahepatic biliary tree or pancreaticoduodenectomy. Frozen section biopsy of the bile duct margin is an important step to exclude dysplasia or tumor involvement. Routine excision of extrahepatic biliary tree for intrahepatic lesion is a matter of debate. Kubota et al. [13] have reported no statistically significant difference in overall survival between patients with or without resection of extrahepatic bile duct. Similarly, routine lymph nodal dissection of hepatoduodenal ligament is not necessary in intrahepatic lesion because lymph nodal metastasis is relatively less common than conventional cholangiocarcinoma even in invasive lesion [3]. Rocha et al. [9] suggested regional lymphadenectomy for hilar or distal bile duct tumors but not for intrahepatic lesion.

The overall survival of IPNB is better than conventional cholangiocarcinoma [12–14]. Lee et al. [12] reported 81 % 5 years survival after curative resection. Similarly, Rocha et al. [9] have shown median survival of 82 months after R0 resection compared to 36 months with R1 resection. Long-term survival depends upon three important factors like depth and percentage of invasive component, cellular type of IPNB, and margins of oncologic resection. Rocha et al. [9] have shown that depth of invasion ≥5 mm and invasive component of ≥10 % were associated with significantly diminished survival. Kim et al. [4] have reported that

histologic subtypes of IPNB are associated with different clinicopathologic features and prognosis. Pancreatobiliary subtype is associated higher histologic grade, more invasive component, more lymph node metastasis, and worse clinical outcomes [4,13]. In addition, patients with mucinous carcinoma showed a better prognosis than patients with tubular adenocarcinoma. Jung et al. [11] have demonstrated that R1 resection was the only prognostic factor for tumor recurrence and overall survival. Similar results were published by Rocha et al. [9] where R1 resection was associated with poor long-term survival (36 months vs 82 months, $p \leq 0.04$). The recurrence rate after surgical resection is about 20 % in benign IPNB and 60 % for malignant cases [4,9]. Most of the recurrences are loco-regional. This high rate of recurrence may be due to incomplete preoperative assessment of the extent of IPNB. Small papillary lesions may not be detected on conventional imaging. These undetected lesions which are usually remote from the main tumor, may be the source of recurrence. Another important factor for recurrence may be the margin positive resection (R1). Therefore, for better outcome, meticulous preoperative assessment of the extent of the tumor, R0 resection and regular follow-up of every 3 to 6 months are recommended.

The distinctive feature of our case is the complexity of its diagnosis. Initial, imaging's failed to provide precise nature of the lesion. Endoscopist misinterpreted mucin expression from papilla as pus because the patient had cholangitis. Repeat CT scan and MRI gave some clue for the diagnosis of IPNB. Exploratory laparotomy and histopathological examinations were needed to confirm the nature of the lesion. We report this case to enhance the awareness that features of biliary obstruction and cystic lesion in relation to bile duct with or without mural nodule, IPNB may be a possibility.

4. Conclusion

IPNB should be considered when a patient present with symptoms of biliary obstruction accompanied by cystic dilatation of the bile duct with wall nodules or papillary mass or solid-cystic mass communicating with the bile duct and the upstream and downstream dilatation of the bile duct. Every attempt should be made to obtain a R0 resection for better long-term survival of the patient.

Consent

Written informed consent was obtained from the patient for publication of the case and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

As per the rules of our Institutional Ethics Committee, ethical approval is not required in our institution to publish anonymous case reports.

Funding

None.

Author contribution

Sukanta Ray: Acquisition of the data, drafting the manuscript, final approval of the version to be submitted.

Arkadeep Dhali: Acquisition of the data, drafting the manuscript, final approval of the version to be submitted.

Hemabha Saha: Conception, design of the study, acquisition of the data, drafting the manuscript, and final approval of the version to be submitted.

Ranajoy Ghosh: Conception, design of the study, acquisition of the data, drafting the manuscript, and final approval of the version to be submitted.

Sujan Khamrui: Acquisition of the data, final approval of the version to be submitted.

Gopal Krishna Dhali: Acquisition of the data, final approval of the version to be submitted.

Research registration number

Not applicable.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Conflict of interest statement

None.

Acknowledgment

Nil.

References

[1] Ohtsuka M, Shimizu H, Kato A, Yoshitomi H, Furukawa K, Tsuyuguchi T, Sakai Y, Yokosuka O, Miyazaki M. Intraductal papillary neoplasms of the bile duct. Int J Hepatol. 2014;2014:459091. doi: https://doi.org/10.1155/2014/459091. Epub 2014 May 18. PMID: 24949206; PMCID: PMC4052179.

[2] Nakanuma Y, Basturk O, Esposito I, Klimstra DS, Komuta M, Zen Y. Intraductal papillary neoplasm of the bile ducts. In: WHOClassification of Tumours. Digestive System Tumours, fifth ed. Lyon:IARC (2019). p. 279–82.

[3] X.S. Wan, Y.Y. Xu, J.Y. Qian, X.B. Yang, A.Q. Wang, L. He, H.T. Zhao, X.T. Sang, Intraductal papillary neoplasm of the bile duct, World J. Gastroenterol. 19 (46) (Dec 14 2013) 8595–8604, https://doi.org/10.3748/wjg.v19.i46.8595 (PMID: 24379576; PMCID: PMC3870504).

[4] K.M. Kim, J.K. Lee, J.U. Shin, K.H. Lee, K.T. Lee, J.Y. Sung, K.T. Jang, J.S. Heo, S. H. Choi, D.W. Choi, J.H. Lim, Clinicopathologic features of intraductal papillary neoplasm of the bile duct according to histologic subtype, Am. J. Gastroenterol. 107 (1) (Jan 2012) 118–125, https://doi.org/10.1038/ajg.2011.316 (Epub 2011 Sep 27. PMID: 21946282).

[5] Y. Tan, C. Milikowski, Y. Toribio, A. Singer, C.P. Rojas, M.T. Garcia-Buitrago, Intraductal papillary neoplasm of the bile ducts: a case report and literature review, World J. Gastroenterol. 21 (43) (Nov 21 2015) 12498–12504, https://doi.org/10.3748/wjg.v21.i43.12498 (PMID: 26604656; PMCID: PMC4649132).

[6] Nakanuma Y, Sato Y, Ojima H, Kanai Y, Aishima S, Yamamoto M, Ariizumi S, Furukawa T, Hayashi H, Unno M, Ohta T; Hepatolithiasis subdivision of intractable hepatobiliary diseases study group of Japan (Chairman, Hirohito Tsubouchi). Clinicopathological characterization of so-called "cholangiocarcinoma with intraductal papillary growth" with respect to "intraductal papillary neoplasm of bile duct (IPNB)". Int J Clin Exp Pathol. 2014 May 15;7(6):3112–22. PMID: 25031730; PMCID: PMC4097227.

[7] I. Fuente, M. Gonzalez, M. de Santibañes, J. Pekolj, O. Mazza, E. de Santibañes, Claria R. Sanchez, Intraductal papillary neoplasm of the bile duct (IPNB): case report and literature review of a challenging disease to diagnose, J. Gastrointest. Cancer 50 (3) (Sep 2019) 578–582, https://doi.org/10.1007/s12029-018-0057-8 (PMID: 29374338).

[8] H.J. Park, S.Y. Kim, H.J. Kim, S.S. Lee, G.S. Hong, J.H. Byun, S.M. Hong, M.G. Lee, Intraductal papillary neoplasm of the bile duct: clinical, imaging, and pathologic features, AJR Am. J. Roentgenol. 211 (1) (Jul 2018) 67–75, https://doi.org/10.2214/AJR.17.19261 (Epub 2018 Apr 9. PMID: 29629808).

[9] F.G. Rocha, H. Lee, N. Katabi, R.P. DeMatteo, Y. Fong, M.I. D'Angelica, P.J. Allen, D.S. Klimstra, W.R. Jarnagin, Intraductal papillary neoplasm of the bile duct: a biliary equivalent to intraductal papillary mucinous neoplasm of the pancreas? Hepatology 56 (4) (Oct 2012) 1352–1360, https://doi.org/10.1002/hep.25786 (Epub 2012 Aug 27. PMID: 22504729).

[10] J.J. Kloek, N.A. van der Gaag, D. Erdogan, E.A. Rauws, O.R. Busch, D.J. Gouma, F. J. ten Kate, T.M. van Gulik, A comparative study of intraductal papillary neoplasia of the biliary tract and pancreas, Hum. Pathol. 42 (6) (Jun 2011) 824–832, https://doi.org/10.1016/j.humpath.2010.09.017 (Epub 2011 Feb 2. PMID: 21292296).

[11] G. Jung, K.M. Park, S.S. Lee, E. Yu, S.M. Hong, J. Kim, Long-term clinical outcome of the surgically resected intraductal papillary neoplasm of the bile duct, J. Hepatol. 57 (4) (Oct 2012) 787–793, https://doi.org/10.1016/j.jhep.2012.05.008 (Epub 2012 May 23. PMID: 22634127).

[12] S.S. Lee, M.H. Kim, S.K. Lee, S.J. Jang, M.H. Song, K.P. Kim, H.J. Kim, D.W. Seo, D. E. Song, E. Yu, S.G. Lee, Y.I. Min, Clinicopathologic review of 58 patients with biliary papillomatosis, Cancer 100 (4) (Feb 15 2004) 783–793, https://doi.org/10.1002/cncr.20031 (PMID: 14770435).

[13] K. Kubota, Y. Nakanuma, F. Kondo, H. Hachiya, M. Miyazaki, M. Nagino, M. Yamamoto, H. Isayama, M. Tabata, H. Kinoshita, T. Kamisawa, K. Inui, Clinicopathological features and prognosis of mucin-producing bile duct tumor and mucinous cystic tumor of the liver: a multi-institutional study by the Japan Biliary Association, J. Hepatobiliary Pancreat. Sci. 21 (3) (Mar 2014) 176–185, https://doi.org/10.1002/jhbp.23 (Epub 2013 Aug 1. PMID: 23908126).

[14] .Baton O, Azoulay D, Adam DV, Castaing D. Major hepatectomy for hilar cholangiocarcinoma type 3 and 4: prognostic factors and longterm outcomes. J Am Coll Surg. 2007 Feb;204(2):250–60. doi: https://doi.org/10.1016/j.jamcollsurg.2006.10.028. Epub 2006 Dec 27. Erratum in: J Am Coll Surg. 2007 Jun;204(6):1304. PMID: 17254929.

[15] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. Lond. Engl. 109 (5) (2023) 1136.

[16] V. Chappet, Cancer epithelial primitif du canal choledoque, Lyon Méd. 76 (1894) 145–157.

[17] R.D. Neumann, V.A. LiVolsi, N.S. Rosenthal, M. Burrell, T.J. Ball, Adenocarcinoma in biliary papillomatosis, Gastroenterology 70 (5 PT.1) (May 1976) 779–782 (PMID: 1261773).

[18] Y. Zen, T. Fujii, K. Itatsu, K. Nakamura, H. Minato, S. Kasashima, H. Kurumaya, K. Katayanagi, A. Kawashima, S. Masuda, H. Niwa, T. Mitsui, Y. Asada, S. Miura, T. Ohta, Y. Nakanuma, Biliary papillary tumors share pathological features with intraductal papillary mucinous neoplasm of the pancreas, Hepatology 44 (5) (Nov 2006) 1333–1343, https://doi.org/10.1002/hep.21387 (PMID: 17058219).

[19] F.T. Bosman, F. Carneiro, R.H. Hruban, N.D. Theise, WHO Classification of Tumours of the Digestive System, World Health Organization, 2010.

[20] L. Barbier, R. Souche, K. Slim, P. Ah-Soune, Long-term consequences of bile duct injury after cholecystectomy, J. Visc. Surg. 151 (4) (Sep 2014) 269–279, https://doi.org/10.1016/j.jviscsurg.2014.05.006 (Epub 2014 Jun 19. PMID: 24954866).

[21] C.J. Kelley, I.S. Benjamin, L.H. Blumgart, Portal hypertension and post-cholecystectomy biliary strictures, Dig. Surg. 3 (4) (1986) 292–296.

[22] T.S. Yeh, J.H. Tseng, T.C. Chen, N.J. Liu, C.T. Chiu, Y.Y. Jan, M.F. Chen, Characterization of intrahepatic cholangiocarcinoma of the intraductal growth-type and its precursor lesions, Hepatology 42 (3) (Sep 2005) 657–664, https://doi.org/10.1002/hep.20837 (PMID: 16116640).

[23] B. Li, Z. Liu, Z. Meng, M. Li, W. Tian, Q. Liu, Surgical treatment of intraductal papillary neoplasm of the bile duct: a report of two cases and review of the literature, Front. Oncol. 12 (Jun 23 2022) 916457, https://doi.org/10.3389/fonc.2022.916457 (PMID: 35814451; PMCID: PMC9259795).

[24] P. Apostolopoulos, K.A. Ekmektzoglou, K. Paraskeva, K. Dimopoulos, K. Paparaskeva, G. Alexandrakis, Intraductal papillary neoplasm of the bile duct: case report and review of the literature, Acta Gastroenterol. Belg. Jan-Mar;81(1): 97-99 (2018) (PMID: 29562383).

[25] Y. Sakai, M. Ohtsuka, H. Sugiyama, R. Mikata, S. Yasui, I. Ohno, Y. Iino, J. Kato, T. Tsuyuguchi, N. Kato, Current status of diagnosis and therapy for intraductal papillary neoplasm of the bile duct, World J. Gastroenterol. 27 (15) (Apr 21 2021) 1569–1577, https://doi.org/10.3748/wjg.v27.i15.1569 (PMID: 33958844; PMCID: PMC8058653).

[26] K. Takanami, T. Yamada, M. Tsuda, K. Takase, K. Ishida, Y. Nakamura, A. Kanno, T. Shimosegawa, M. Unno, S. Takahashi, Intraductal papillary mucininous neoplasm of the bile ducts: multimodality assessment with pathologic correlation, Abdom. Imaging 36 (4) (Aug 2011) 447–456, https://doi.org/10.1007/s00261-010-9649-x (PMID: 20959978).