**Name of Journal:** *World Journal of Clinical Cases*

**Manuscript NO:** 88044

**Manuscript Type:** CASE REPORT

**Surgically treating a rare and asymptomatic intraductal papillary neoplasm of the bile duct: A case report**

Zhu SZ *et al*. Surgical treatment of IPNB

Shen-Zhen Zhu, Zhao-Feng Gao, Xiao-Rong Liu, Xiao-Guang Wang, Fei Chen

**Shen-Zhen Zhu,** Department of General Surgery, Jiaxing Second Hospital, Jiaxing 314000, Zhejiang Province, China

**Zhao-Feng Gao, Xiao-Rong Liu, Xiao-Guang Wang, Fei Chen,** Department of Hepatobiliary Surgery, Jiaxing Second Hospital, Jiaxing 314000, Zhejiang Province, China

**Co-corresponding authors:** Xiao-Guang Wang and Fei Chen.

**Author contributions:** Gao ZF contributed to the treatment of cases and provided figures; Zhu SZ researched the data and wrote the manuscript; Liu XR contributed to the discussion; Chen F and Wang XG guided the writing ideas and reviewed the manuscript. All authors have read and approved the final version of the manuscript. Wang XG and Chen F contributed equally to this work as co-corresponding authors. The reasons for designating Wang XG and Chen F as co-corresponding authors are threefold. First, they provided great insights throughout the revision and improvement of the manuscript. Second, they helped the team to obtain support from relevant funds. Third, they also rationalized the assignment of tasks to our whole team, and the selection of them as co-corresponding authors recognizes and respects their equal contributions and commends the collaborative spirit of our research team. All the members agreed to designate Wang XG and Chen F as the co-corresponding authors.

**Supported by** Zhejiang Provincial Natural Science Foundation of China Under Grant, No. LY21H160046.

**Corresponding author: Xiao-Guang Wang, Doctor, Academic Editor, Doctor,** Department of Hepatobiliary Surgery, Jiaxing Second Hospital, No. 1518 Huancheng North Road, Nanhu District, Jiaxing 314000, Zhejiang Province, China. xiaoguangwangs@163.com

**Received:** September 7, 2023

**Revised:** November 28, 2023

**Accepted:** December 26, 2023

**Published online:**

**Abstract**

BACKGROUND

Intraductal papillary neoplasms of the bile duct (IPNBs) are rare and characterized by papillary growth within the bile duct lumen. IPNB is similar to obstructive biliary pathology. In this report, we present an unexpected case of asymptomatic IPNB and consolidate our findings with the relevant literature to augment our understanding of this condition. Integrating relevant literature contributes to a more comprehensive understanding of the disease.

CASE SUMMARY

A 66-year-old Chinese male patient was admitted to our hospital for surgical intervention after gallstones were discovered during a routine physical examination. Preoperative imaging revealed a lesion on the left side of the liver, which raised the suspicion of IPNB. A laparoscopic left hemihepatectomy was performed, and subsequent histopathological examination confirmed the diagnosis of IPNB. At the 3-mo postoperative follow-up, the patient reported good recovery and no metastasis. IPNB can manifest both latently and asymptomatically. Radical surgical resection is the most effective treatment for IPNB.

CONCLUSION

Hepatic and biliary masses, should be considered to diagnose IPNB. Prompt surgery and vigilant follow-up are crucial in determining prognosis.

**Key Words:** Intraductal papillary neoplasm of the bile duct; Tumor; Surgical treatment; Prognosis; Case report

Zhu SZ, Gao ZF, Liu XR, Wang XG, Chen F. Surgically treating a rare and asymptomatic intraductal papillary neoplasm of the bile duct: A case report. *World J Clin Cases* 2023; In press

**Core Tip:** Intraductal papillary neoplasms are relatively uncommon in clinical practice, often eluding detection by clinicians owing to the inadequacy of conventional imaging tests such as abdominal ultrasound and computed tomography, to identify and diagnose tumors. In the present case, the patient was asymptomatic. However, if the tumor was not detected and resected promptly, the patient could have lost the opportunity for surgical intervention when developing relevant biliary symptoms. Early detection and surgical intervention hold promise for a positive prognosis. Clinicians must further enhance their understanding of the clinical attributes and imaging indications to prevent missed diagnoses.

**INTRODUCTION**

Intraductal papillary neoplasms of the bile duct (IPNBs) are rare tumors that predominantly affect middle-aged and elderly individuals. It exhibits a slightly higher prevalence in men than in women and has significant potential for malignancy. Most patients present with symptoms, such as epigastric pain, acute cholangitis, and jaundice, accompanied by elevated liver enzyme levels. However, asymptomatic cases are less frequent. Here, we report a case of asymptomatic IPNB and conduct a comprehensive review of the relevant literature to enhance our understanding of this disease.

**CASE PRESENTATION**

***Chief complaints***

A 66-year-old male patient was admitted to our hospital with a 7-year history of gallbladder stones, detected during a physical examination, leading to a proposed laparoscopic cholecystectomy.

***History of present illness***

Preoperative abdominal ultrasonography revealed multiple gallstones and cysts in the left lobe of the liver. Plain computed tomography (CT) of the chest and abdomen revealed low-density lesions in the left hepatic lobe, prompting the recommendation for an enhanced abdominal CT examination.

***History of past illness***

The patient had no prior history of surgery but had a history of multiple chronic conditions, including hypertension, diabetes, hyperlipidemia, and atrial fibrillation. The patient consistently adhered to the medication regimen and reported satisfactory management.

***Personal and family history***

The patient denied any family history of tumors.

***Physical examination***

The patient was alert and in good condition. No abnormal symptoms such as jaundice, nausea, vomiting, or acid reflux were noted. An abdominal examination revealed no tenderness, rebound tenderness, or muscle tension. No abnormal masses were palpable, and the Murphy's sign was negative.

***Laboratory examinations***

Total bilirubin, 20.6 μmol/L (reference range: 0-23 μmol/L); direct bilirubin, 7 μmol/L (reference range: 0-4 μmol/L); indirect bilirubin, 13.6 μmol/L (reference range: 0-17 μmol/L); alanine transaminase (ALT), 13 U/L (reference range: 9-50 U/L); aspartate transaminase (AST), 19 U/L (reference range: 15-40 U/L); carcinoembryonic antigen (CEA), 5.46 ng/mL (reference range: 0-5 ng/mL); alpha fetoprotein, 2.61 ng/mL (reference range: 0-20 ng/mL); carbohydrate antigen 199 (CA199), 29.88 U/mL (reference range: 0-37 U/mL). The rest of the laboratory results were normal. Pathological examination of the lesion revealed an IPNB with severe epithelial dysplasia or high-grade intraepithelial neoplasia (Figure 1).

***Imaging examinations***

Abdominal enhanced CT arterial phase (Figure 2A), abdominal enhanced CT venous phase (Figure 2B), and an initial diagnosis of a cystic, solid lesion in the left lobe of the liver with dilatation of the distal intrahepatic bile ducts were made; cholangiocarpal cystadenoma was ruled out, and magnetic resonance (MR) enhancement examination was recommended. Subsequently, abdominal MR imaging (MRI) + MR cholangiopancreatography (MRCP) (enhanced) examination (Figure 2C-E) confirmed the presence of cystic and solid lesions in the left hepatic lobe, along with dilation of the left intrahepatic bile duct, indicative of IPNB.

**FINAL DIAGNOSIS**

The patient was diagnosed with IPNB, accompanied by severe epithelial dysplasia and high-grade intraepithelial neoplasia.

**TREATMENT**

The patient underwent a laparoscopic left hemihepatectomy and cholecystectomy. No additional masses were identified perioperatively, and no subsequent treatment was required.

**OUTCOME AND FOLLOW-UP**

The patient exhibited favorable postoperative recovery and underwent regular follow-ups at our outpatient clinic for three months using abdominal CT, with no evidence of metastasis or recurrence.

**DISCUSSION**

According to the latest WHO classification, IPNB, recognized as a precancerous lesion in cholangiocarcinoma (CCA), is predominantly characterized by papillary or villous growth within the bile duct lumen[1]. More than 30% of patients with IPNB exhibit significant mucus secretion into the lumen[2]. The pathogenesis of IPNB remains unclear; however, investigations have identified major risk factors, including hepatic bile duct stones, hepatic schistosome infection, primary sclerosing cholangitis, congenital biliary anomalies, and exposure to substances, such as chlorinated organic solvents. Other contributing factors include bile duct malformations, familial adenomatous polyposis, and Gardner's syndrome. The progression of IPNB is gradual, and is initiated by inflammation due to biliary stasis, biliary infections, and CCAs, ultimately culminating in a multistage transformation of the bile ducts, marked by proliferation, heterotopic hyperplasia, and carcinoma development[3]. Molecular studies have identified mutations in CTNNB1, STK11, and GNAS in patients with IPNB concurrently with intraductal papillary mucinous neoplasm (IPMN)[4]. Kirsten rat sarcoma viral oncogene mutations are risk factors for IPNB progression and offer potential diagnostic and therapeutic avenues[5]. Tanaka *et al*[6] identified EVI1 expression in IPNB, highlighting its potential as a prognostic marker. Notably, patients with MUC1-expressing IPNB tumors have a more unfavorable prognosis[7]. Owing to the relatively subtle clinical symptoms of IPNB compared with those of other tumors, a significant number of patients with IPNB are diagnosed after the disease has advanced, leading to bile duct obstruction. Subsequently, these patients gradually develop associated biliary symptoms, including jaundice, abdominal pain, and cholangitis[8]. The current study's findings indicate a higher prevalence of IPNB in East Asia, particularly in regions such as Japan, Korea, and Thailand, where intrahepatic bile duct stones are prevalent[9]. IPNB is also considered the biliary counterparts of IPMNs of the pancreas because of their mucin-hypersecretory properties[10]. To enhance staging and prognostic accuracy, IPNB has been further classified into four distinct subtypes: Pancreaticobiliary, intestinal, eosinophilic, and gastric; the intestinal subtype is the most prevalent and displays the highest incidence of malignancy. Currently, the commonly used modified IPNB staging systems include types 1 (intrahepatic biliary) and 2 (extrahepatic biliary). Approximately 40% of IPNB cases were classified as type 1, and 60% as type 2. Notably, mucin hypersecretion is more prevalent in type 1 IPNB (I-IPNB) compared to type 2[11,12]. Recent studies have demonstrated that the modified two-tier grading system effectively reflects postoperative survival compared to the traditional grading approach, contributing to a comprehensive postoperative assessment of IPNB[13]. Additionally, one research team discovered that the survival rate for type 1 was notably superior to that for type 2 by employing a scoring system encompassing six pathological characteristics (such as location, mucin secretion, and tissue structure)[14]. Compared with CCA, IPNB generally exhibits a more favorable overall prognosis. However, given the potential for malignant transformation, early diagnosis and prompt treatment of IPNB remain central to ongoing research. Our case was incidentally discovered when the patient was admitted to the hospital for surgical treatment of gallbladder stones. The majority of cases were identified as a consequence of disease progression, presenting with biliary symptoms that led them to seek medical attention for laboratory and imaging assessments. Nonetheless, routine laboratory tests, such as ALT/AST/alkaline phosphatase and tumor markers CEA/CA199, lack distinct specificity and often exhibit elevated levels in many patients with typical biliary inflammation[15]. CT and MRI are remain the primary diagnostic tools for imaging evaluation. However, patients with IPNB often present with concurrent biliary inflammation and stones, resulting in a low rate of early clinical diagnosis. Currently, choledochoscopy should be employed in conjunction with CT and MRI for the precise localization of IPNB. Transoral cholangioscopy (POCS) enables the assessment of the scope of bile duct lesions, thereby facilitating the formulation of a tailored surgical strategy[16]. In IPNB cases characterized by substantial mucin secretion, percutaneous transhepatic cholangioscopy (PTCS) offers greater advantages than POCS[17]. The 2012 guidelines identified pertinent predictors, such as obstructive jaundice, duct size, wall nodules, and abrupt size changes, as potential indicators of the extent of malignancy in IPNB[18]. A recent study conducted by a Korean research team revealed that of the 116 cases of I-IPNB, 62 (53.4%) exhibited invasive carcinomas, whereas 61 (76.3%) of the 80 cases of type 2 extrahepatic IPNB displayed invasive carcinomas. Multifactorial analysis indicated that mural nodules of < 12 mm and intensified mural nodules were predictors of malignancy in I-IPNB and type 2 IPNB, respectively. In addition, 43.7% of patients with nonsurgical mucosal alloplastic hyperplasia would develop malignancies within three years[19].

Prompt initiation of surgical treatment is imperative for patients diagnosed with IPNB, likely achieving complete margin-negative (R0), significantly enhancing patient prognosis[20]. Conventional surgical approaches include partial hepatectomy, choledochotomy, pancreaticoduodenectomy, hepatopancreaticoduodenectomy, and liver transplantation. The optimal surgical choice is determined through preoperative multidisciplinary team discussions, PTCS, and POCS integrated with intraoperative cholangioscopy. Recently, a Japanese research team effectively employed red dichroic imaging technology for precise tumor extent assessment during preoperative POCS in a 75-year-old IPNB patient, consequently facilitating a successful pancreaticoduodenectomy[21]. Palliative modalities are frequently employed in patients with severe preoperative bile duct inflammation or in advanced cases that are not amenable to surgical intervention. These include endoscopic nasobiliary drainage and endoscopic retrograde cholangiopancreatography biliary stenting aimed to mitigate the patient's symptoms. Innovative techniques, such as radiofrequency ablation and photodynamic therapy have emerged as viable treatment alternatives[22,23]. A Korean research team successfully alleviated jaundice in a patient with advanced IPNB using argon plasma coagulation, and the current follow-up period extends beyond two years[24]. Advancements in novel technologies have the potential to enhance patient survival significantly. Despite IPNB's notably improved overall prognosis compared with CCA, with a 5-year survival rate exceeding 80%[25], ongoing regular follow-up remains imperative. Notably, approximately 13%-29% of patients with surgically resected IPNB experience recurrence within a short timeframe. This recurrence rate escalates to 47%-62% in cases where the patient is diagnosed with invasive IPNB[26].

**CONCLUSION**

In conclusion, IPNB, recognized as a precancerous lesion, has a favorable overall prognosis. However, the precise pathogenesis and mechanisms underlying IPNB progression remain unclear. Furthermore, nonspecific clinical manifestations pose a challenge for early diagnosis. Meticulous preoperative imaging, intraoperative cholangioscopy, and prompt pathological evaluation of resection margins are crucial for the informed selection of an appropriate surgical strategy. A comprehensive exploration of molecular targeted therapy coupled with vigilant postoperative monitoring will aid in formulating a more informed and rational treatment protocol for individuals affected by IPNB.

**ACKNOWLEDGEMENTS**

We extend our gratitude to the patients for their valuable contributions to this case report. Our appreciation also goes to the Department of General Surgery at the Second Affiliated Hospital of Jiaxing University for their support in managing this case.

**REFERENCES**

1 **Nagtegaal ID**, Odze RD, Klimstra D, Paradis V, Rugge M, Schirmacher P, Washington KM, Carneiro F, Cree IA; WHO Classification of Tumours Editorial Board. The 2019 WHO classification of tumours of the digestive system. *Histopathology* 2020; **76**: 182-188 [PMID: 31433515 DOI: 10.1111/his.13975]

2 **Wang X**, Cai YQ, Chen YH, Liu XB. Biliary tract intraductal papillary mucinous neoplasm: report of 19 cases. *World J Gastroenterol* 2015; **21**: 4261-4267 [PMID: 25892877 DOI: 10.3748/wjg.v21.i14.4261]

3 **Nakanuma Y**, Kakuda Y, Sugino T, Sato Y, Fukumura Y. Pathologies of Precursor Lesions of Biliary Tract Carcinoma. *Cancers (Basel)* 2022; **14** [PMID: 36358777 DOI: 10.3390/cancers14215358]

4 **Zen Y**, Akita M. Neoplastic Progression in Intraductal Papillary Neoplasm of the Bile Duct. *Arch Pathol Lab Med* 2023 [PMID: 36800543 DOI: 10.5858/arpa.2022-0407-RA]

5 **Sun T**, Zuo T, Hui P, Cai G. Significance of KRAS mutation testing in biliary brushing cytology specimens: A 10-year retrospective review. *Cancer Cytopathol* 2022; **130**: 558-565 [PMID: 35417072 DOI: 10.1002/cncy.22579]

6 **Tanaka M**, Shibahara J, Ishikawa S, Ushiku T, Morikawa T, Shinozaki-Ushiku A, Hayashi A, Misumi K, Tanaka A, Katoh H, Sakuma K, Kokudo T, Inagaki Y, Arita J, Sakamoto Y, Hasegawa K, Fukayama M. EVI1 expression is associated with aggressive behavior in intrahepatic cholangiocarcinoma. *Virchows Arch* 2019; **474**: 39-46 [PMID: 30349952 DOI: 10.1007/s00428-018-2476-0]

7 **Bennett S**, Marginean EC, Paquin-Gobeil M, Wasserman J, Weaver J, Mimeault R, Balaa FK, Martel G. Clinical and pathological features of intraductal papillary neoplasm of the biliary tract and gallbladder. *HPB (Oxford)* 2015; **17**: 811-818 [PMID: 26278323 DOI: 10.1111/hpb.12460]

8 **Lee SS**, Kim MH, Lee SK, Jang SJ, Song MH, Kim KP, Kim HJ, Seo DW, Song DE, Yu E, Lee SG, Min YI. Clinicopathologic review of 58 patients with biliary papillomatosis. *Cancer* 2004; **100**: 783-793 [PMID: 14770435 DOI: 10.1002/cncr.20031]

9 **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 [PMID: 24949206 DOI: 10.1155/2014/459091]

10 **Kubota K**, Nakanuma Y, Kondo F, Hachiya H, Miyazaki M, Nagino M, Yamamoto M, Isayama H, Tabata M, Kinoshita H, Kamisawa T, Inui K. 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* 2014; **21**: 176-185 [PMID: 23908126 DOI: 10.1002/jhbp.23]

11 **Kubota K**, Jang JY, Nakanuma Y, Jang KT, Haruyama Y, Fukushima N, Furukawa T, Hong SM, Sakuraoka Y, Kim H, Matsumoto T, Lee KB, Zen Y, Kim J, Miyazaki M, Choi DW, Heo JS, Endo I, Hwang S, Nakamura M, Han HS, Uemoto S, Park SJ, Hong EK, Nanashima A, Kim DS, Kim JY, Ohta T, Kang KJ, Fukumoto T, Nah YW, Seo HI, Inui K, Yoon DS, Unno M. Clinicopathological characteristics of intraductal papillary neoplasm of the bile duct: a Japan-Korea collaborative study. *J Hepatobiliary Pancreat Sci* 2020; **27**: 581-597 [PMID: 32511838 DOI: 10.1002/jhbp.785]

12 **Nakanuma Y**, Uesaka K, Okamura Y, Terada T, Fukumura Y, Kakuda Y, Sugino T, Sato Y, Taek JK, Park YN. Reappraisal of pathological features of intraductal papillary neoplasm of bile duct with respect to the type 1 and 2 subclassifications. *Hum Pathol* 2021; **111**: 21-35 [PMID: 33508254 DOI: 10.1016/j.humpath.2021.01.002]

13 **Nakanuma Y**, Jang KT, Fukushima N, Furukawa T, Hong SM, Kim H, Lee KB, Zen Y, Jang JY, Kubota K. A statement by the Japan-Korea expert pathologists for future clinicopathological and molecular analyses toward consensus building of intraductal papillary neoplasm of the bile duct through several opinions at the present stage. *J Hepatobiliary Pancreat Sci* 2018; **25**: 181-187 [PMID: 29272078 DOI: 10.1002/jhbp.532]

14 **Onoe S**, Ebata T, Yokoyama Y, Igami T, Mizuno T, Yamaguchi J, Watanabe N, Otsuka S, Nakamura S, Shimoyama Y, Nagino M. A clinicopathological reappraisal of intraductal papillary neoplasm of the bile duct (IPNB): a continuous spectrum with papillary cholangiocarcinoma in 181 curatively resected cases. *HPB (Oxford)* 2021; **23**: 1525-1532 [PMID: 33832834 DOI: 10.1016/j.hpb.2021.03.004]

15 **Jung G**, Park KM, Lee SS, Yu E, Hong SM, Kim J. Long-term clinical outcome of the surgically resected intraductal papillary neoplasm of the bile duct. *J Hepatol* 2012; **57**: 787-793 [PMID: 22634127 DOI: 10.1016/j.jhep.2012.05.008]

16 **Tringali A**, Milluzzo SM, Ardito F, Laurenzi A, Ettorre GM, Barbaro B, Ricci R, Giuliante F, Boškoski I, Costamagna G. Peroral-cholangioscopy to plan surgery for protruding biliary lesions: report of four cases. *Ther Adv Gastrointest Endosc* 2022; **15**: 26317745221139735 [PMID: 36465430 DOI: 10.1177/26317745221139735]

17 **Tsuyuguchi T**, Sakai Y, Sugiyama H, Miyakawa K, Ishihara T, Ohtsuka M, Miyazaki M, Yokosuka O. Endoscopic diagnosis of intraductal papillary mucinous neoplasm of the bile duct. *J Hepatobiliary Pancreat Sci* 2010; **17**: 230-235 [PMID: 19669677 DOI: 10.1007/s00534-009-0153-z]

18 **Tanaka M**, Fernández-del Castillo C, Adsay V, Chari S, Falconi M, Jang JY, Kimura W, Levy P, Pitman MB, Schmidt CM, Shimizu M, Wolfgang CL, Yamaguchi K, Yamao K; International Association of Pancreatology. International consensus guidelines 2012 for the management of IPMN and MCN of the pancreas. *Pancreatology* 2012; **12**: 183-197 [PMID: 22687371 DOI: 10.1016/j.pan.2012.04.004]

19 **Han SY**, Kim DU, Nam HS, Kang DH, Jang SI, Lee DK, Shin DW, Cho KB, Yang MJ, Hwang JC, Kim JH, So H, Bang SJ, Sung MJ, Kwon CI, Lee DW, Cho CM, Cho JH. Comparison of the Malignant Predictors in Intrahepatic and Extrahepatic Intraductal Papillary Neoplasm of the Bile Duct. *J Clin Med* 2022; **11** [PMID: 35407592 DOI: 10.3390/jcm11071985]

20 **Chan WH**, Chen CM, Wang SY, Wu RC, Chen TC, Lee HK, Lin CH, Yeh CN. Intraductal papillary neoplasm of the bile duct presenting with hepatogastric fistula: a case report and literature review. *Front Oncol* 2023; **13**: 1193918 [PMID: 37274235 DOI: 10.3389/fonc.2023.1193918]

21 **Koiwai A**, Hirota M, Murakami K, Katayama T, Kin R, Endo K, Kogure T, Takasu A, Sakurai H, Kondo N, Takami K, Yamamoto K, Katayose Y, Satoh K. Direct peroral cholangioscopy with red dichromatic imaging 3 detected the perihilar margin of superficial papillary extension in a patient with intraductal papillary neoplasm of the bile duct. *DEN Open* 2023; **3**: e228 [PMID: 36998349 DOI: 10.1002/deo2.228]

22 **Natov NS**, Horton LC, Hegde SR. Successful endoscopic treatment of an intraductal papillary neoplasm of the bile duct. *World J Gastrointest Endosc* 2017; **9**: 238-242 [PMID: 28572878 DOI: 10.4253/wjge.v9.i5.238]

23 **Bechmann LP**, Hilgard P, Frilling A, Schumacher B, Baba HA, Gerken G, Zoepf T. Successful photodynamic therapy for biliary papillomatosis: a case report. *World J Gastroenterol* 2008; **14**: 4234-4237 [PMID: 18636672 DOI: 10.3748/wjg.14.4234]

24 **Arai J**, Kato J, Toda N, Kurokawa K, Shibata C, Kurosaki S, Funato K, Kondo M, Takagi K, Kojima K, Ohki M, Seki M, Tagawa K. Long-term survival after palliative argon plasma coagulation for intraductal papillary mucinous neoplasm of the bile duct. *Clin J Gastroenterol* 2021; **14**: 314-318 [PMID: 32779145 DOI: 10.1007/s12328-020-01199-0]

25 **Kim JR**, Jang KT, Jang JY, Lee K, Kim JH, Kim H, Kim SW, Kwon W, Choi DW, Heo J, Han IW, Hwang S, Kim WJ, Hong SM, Kim DS, Yu YD, Kim JY, Nah YW, Park HW, Choi HJ, Han HS, Yoon YS, Park SJ, Hong EK, Seo HI, Park DY, Kang KJ, Kang YN, Yu HC, Moon WS, Lim CS, Bae JM, Jo S, Lee W, Roh YH, Jeong JS, Jeong CY, Lee JS, Song IS, Kim KH, Kim HG, Cho CH, Joo SH, Won KY, Kim HJ, Choi JH, Chu CW, Lee JH, Park IY, Lee H, Lee SE, Kim HS, Lee HK, Cho MS, Kim H, Han KM. Clinicopathologic analysis of intraductal papillary neoplasm of bile duct: Korean multicenter cohort study. *HPB (Oxford)* 2020; **22**: 1139-1148 [PMID: 31837945 DOI: 10.1016/j.hpb.2019.11.007]

26 **Le A**, Mathew A, Khrais A, Khmelnitsky I, Vossough S. Intraductal Papillary Neoplasm of the Bile Duct: A Rare Disease and Presentation. *Cureus* 2023; **15**: e34556 [PMID: 36879718 DOI: 10.7759/cureus.34556]

**Footnotes**

**Informed consent statement:** Informed written consent was obtained from the patient for publication of this report and any accompanying images.

**Conflict-of-interest statement:** The authors declare that they have no conflict of interest.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Open-Access:** This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: https://creativecommons.org/Licenses/by-nc/4.0/

**Provenance and peer review:** Unsolicited article; Externally peer reviewed.

**Peer-review model:** Single blind

**Peer-review started:** September 7, 2023

**First decision:** November 20, 2023

**Article in press:**

**Specialty type:** Medicine, research and experimental

**Country/Territory of origin:** China

**Peer-review report’s scientific quality classification**

Grade A (Excellent): A

Grade B (Very good): 0

Grade C (Good): 0

Grade D (Fair): 0

Grade E (Poor): 0

**P-Reviewer:** Yildiz K, Turkey **S-Editor:** Qu XL **L-Editor:** A **P-Editor:** Qu XL

**Figure Legends**



**Figure 1 The pathological result suggestive of a paraganglioma (hepatic encephalopathy × 400).**



**Figure 2 Abdominal enhanced computed tomography and magnetic resonance imaging + magnetic resonance cholangiopancreatography (enhanced) examination.** A and B: The initial diagnosis of a cystic solid lesion in the left lobe of the liver with dilatation of the distal intrahepatic bile ducts. Cholangiocarpal cystadenoma was ruled out, and a magnetic resonance enhancement examination was recommended; C-E: Presence of cystic and solid lesions in the left hepatic lobe and dilation of the left intrahepatic bile duct, indicative of intraductal papillary neoplasms of the bile duct.