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ORIGINAL ARTICLE

An observational and comparative study on intraductal papillary mucinous neoplasm of the biliary tract and the pancreas from a Chinese cohort

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Summary

Background: Intraductal papillary mucinous neoplasms of the biliary tract (BT-IPMNs) are unique but very rare biliary tumors. The relationship between BT-IPMNs and intraductal papillary mucinous neoplasm of the pancreas (P-IPMNs) was still unclear and controversial.

Objective: We aimed to evaluate the clinical, radiological, histopathological, and prognostic characteristics of BT-IPMNs and P-IPMNs to achieve a better understanding of these two rare bilio-pancreatic diseases and their connections.

Methods: Data of a total of 116 patients who were all surgically treated and histopathologically diagnosed as BT-IPMNs or P-IPMNs from January 2004 to December 2014 in our single institution was all retrospectively collected and analyzed.

Results: This study respectively enrolled 32 patients with BT-IPMNs and 84 ones with P-IPMNs. The differences between BT-IPMNs and P-IPMNs in age, sex ratios, clinical presentation, elevated tumor markers and proportion of malignancy were not statistically significant ($P > 0.05$), while the tumor diameter of BT-IPMNs was notably smaller than P-IPMNs (1.72 cm, 4.56 cm, respectively; $P = 0.028$). Patients with BT-IPMNs were more likely manifest the symptoms of cholangitis, compared to those with P-IPMNs who showed pancreatitis (75%, 30%, respectively; $P = 0.039$). Bile duct dilatation (100%), tumor of bile duct cavity (50%) or/and cystic dilatation of the bile duct (50%) were the typical manifestations of preoperative imaging examinations of BT-IPMNs, in which tumors were mainly located in intrahepatic or hepatic hilar region (26, 81%). Surgery has been the curable treatment for BT-IPMNs in which left hepatic lobectomy

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was the most commonly performed procedure (20, 63%). Finally, compared with P-IPMNs, the overall mean survival time of patients with BT-IPMNs was a little shorter (59.1 mon, 86.7 mon, respectively; $P = 0.002$).
Conclusion: BT-IPMNs are a sort of rare and separate biliary tract neoplasm, which might be related with the stones of biliary tract or the infections of parasite. Although arising in different organs and representing different features, BT-IPMNs and P-IPMNs shared considerable clinical and pathological similarities which might represent related or similar development process in the bilio-pancreatic duct systems.

Introduction

Arising from biliary ductal epithelium, intraductal papillary mucinous neoplasms of the biliary tract (BT-IPMNs) were characterized by the predominant intraductal papillary growth and/or mucin-secreting lesion, which can occur anywhere along the biliary tree with the dilated bile duct [1]. The growths of BT-IPMNs are sometimes multifocal for their occurring in the intrahepatic and/or extrahepatic bile ducts synchronously. With the potential of evolving from low-grade dysplasia to invasive carcinoma, BT-IPMNs were then added to the 2010 World Health Organization (WHO) classification as a kind of unique biliary neoplasm [2]. In recent years, though still very rare, BT-IPMNs have been increasingly recognized and reported [3—5].

Intraductal papillary mucinous neoplasms of the pancreas (P-IPMNs) are a sort of pancreatic cystic tumors which were also characterized by the intraductal papillary hyperplasia and mucus secretion within dilated pancreatic ducts and ductules [6]. Although they are not uncommon as well as BT-IPMNs, accounting for only 25% of all pancreatic cystic tumors and 16% of all resected pancreatic tumors [7,8], P-IPMNs have been well-documented since they were formally named by WHO in 1996 [9]. As a special pancreatic neoplasm which could present the potential to be malignant, P-IPMNs were widely accepted and have been gradually paid more and more attention to in the past years [10].

Due to the similar characteristics of mucus-secreting papillary lesion originated from ductal epithelium, and the common embryologic origin as a ventral outgrowth of the foregut [11], some studies reported BT-IPMNs had a close relationship with P-IPMNs, and that BT-IPMNs have been recently proposed as the counterpart of P-IPMNs which might be a special manifestation of P-IPMNs in the biliary tract system [5,6,12,13,4,14,15]. However, contrary to P-IPMNs, BT-IPMNs are not well established in the literatures. Despite an increasing number of studies focusing on BT-IPMNs, it was still unclear about several aspects of BT-IPMNs, such as the radiological and histopathological features, surgical strategies and clinical outcome. Meanwhile, it remains controversial whether BT-IPMNs are similar to P-IPMNs in terms of their biological behavior. In the present study, based on the clinical data of our hospital, we carried out a single-institution, retrospective analysis and comparison of patients who were clinically diagnosed as BT-IPMNs and P-IPMNs. We aimed to evaluate the clinical, radiological, histopathological, and prognostic characteristics of BT-IPMNs and P-IPMNs to achieve a better understanding of these two rare bilio-pancreatic diseases.

Materials and methods

Patients selection

This study enrolled respectively 32 and 84 eligible patients at our institution from January 2004 to December 2014, who were all surgically treated and pathologically diagnosed as BT-IPMNs and P-IPMNs, while those with only clinical suspicion but not postoperatively pathological confirmations were not included in our study. The research was approved by the local ethics committee. Data, including patients' demographics (gender and age), clinical presentations at the time of admission, preoperative radiological examinations, surgical information (procedures of operation, intraoperative findings), postoperative detailed pathological diagnosis, etc. were all retrospectively reviewed from the paper-based or electronic medical records. Patients with papillary lesions arising from the gallbladder and ampulla of Vater, the mucinous cystic tumor of liver and other pancreatic cystic tumors were all excluded from this analysis. Remarkably, BT-IPMNs of our study should present either microscopic or macroscopic mucus secretions.

Tumor characteristics

Features of all relevant tumors (such as size, location, quantity, lymph invasion, distant metastasis, surgical margin, etc.) were mainly based on the intraoperative findings and postoperative pathological reports of all resected specimen. BT-IPMNs were histologically defined as the widely accepted pathological criteria of papillary and/or mucinous tumors demonstrating a clear origin from biliary epithelium in the extrahepatic and intrahepatic large bile ducts, with solitary or diffuse intraductal growth and histological spectrum ranging from benign disease to invasive malignancy [16—19]. The neoplasms were recognized as adenoma, borderline tumor, carcinoma in situ, and invasive carcinoma according to the degree of epithelial dysplasia of the WHO classification, which were then artificially divided into benign group (adenoma and borderline tumor) and malignant one (carcinoma in situ and invasive carcinoma) referring to the pathological diagnosis [10].