**Supplement 1. Checklist of items in reporting this systematic review (PRISMA statement)**

|  |  |  |  |
| --- | --- | --- | --- |
| **Section/topic** | **Item** | **Checklist item** | **Reported**  **on page** |
| **Title** | | | |
| Title | 1 | Identify the report as a systematic review, meta-analysis, or both | 1 |
| **Abstract** |  |  |  |
| Structured summary | 2 | Provide a structured summary including, as applicable, background, objectives, data sources, study eligibility criteria, participants, interventions, study appraisal and synthesis methods, results, limitations, conclusions and implications of key findings, systematic review registration number | 2 |
| **Introduction** | | | |
| Rationale | 3 | Describe the rationale for the review in the context of what is already known | 4 |
| Objectives | 4 | Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS) | 5 |
| **Methods** | | | |
| Protocol and registration | 5 | Indicate if a review protocol exists, if and where it can be accessed (such as web address), and, if available, provide registration information including registration number | N/A |
| Eligibility criteria | 6 | Specify study characteristics (such as PICOS, length of follow-up) and report characteristics (such as years considered, language, publication status) used as criteria for eligibility, giving rationale | 5-6 |
| Information sources | 7 | Describe all information sources (such as databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched | 5 |
| Search | 8 | Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated | 5 |
| Study selection | 9 | State the process for selecting studies (that is, screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis | 5-6 |
| Data collection process | 10 | Describe method of data extraction from reports (such as piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators | 6 |
| Data items | 11 | List and define all variables for which data were sought (such as PICOS, funding sources) and any assumptions and simplifications made | 6 |
| Risk of bias in individual studies | 12 | Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis | 7 |
| Summary measures | 13 | State the principal summary measures (such as risk ratio, difference in means) | 7 |
| Synthesis of results | 14 | Describe the methods of handling data and combining results of studies, if done, including measures of consistency (such as I2 statistic) for each meta-analysis | 7 |
| Risk of bias across studies | 15 | Specify any assessment of risk of bias that may affect the cumulative evidence (such as publication bias, selective reporting within studies) | 7 |
| Additional analyses | 16 | Describe methods of additional analyses (such as sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified | N/A |
| **Results** | | | |
| Study selection | 17 | Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram | 8 |
| Study characteristics | 18 | For each study, present characteristics for which data were extracted (such as study size, PICOS, follow-up period) and provide the citations | 9 |
| Risk of bias within studies | 19 | Present data on risk of bias of each study and, if available, any outcome-level assessment (see item 12). | N/A |
| Results of individual studies | 20 | For all outcomes considered (benefits or harms), present for each study (a) simple summary data for each intervention group and (b) effect estimates and confidence intervals, ideally with a forest plot | N/A |
| Synthesis of results | 21 | Present results of each meta-analysis done, including confidence intervals and measures of consistency | N/A |
| Risk of bias across studies | 22 | Present results of any assessment of risk of bias across studies (see item 15) | 15 |
| Additional analysis | 23 | Give results of additional analyses, if done (such as sensitivity or subgroup analyses, meta-regression) (see item 16) | N/A |
| **Discussion** | | | |
| Summary of evidence | 24 | Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (such as health care providers, users, and policy makers | 16 |
| Limitations | 25 | Discuss limitations at study and outcome level (such as risk of bias), and at review level (such as incomplete retrieval of identified research, reporting bias) | 20 |
| Conclusions | 26 | Provide a general interpretation of the results in the context of other evidence, and implications for future research | 20-21 |
| **Funding** | | | |
| Funding | 27 | Describe sources of funding for the systematic review and other support (such as supply of data) and role of funders for the systematic review | 21 |

N/A: not applicable

Modified from: David Moher, Alessandro Liberati, Jennifer Tetzlaff, Douglas G Altman. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. BMJ 2009;339:b2535.

**Supplement 2. Actual search study of this systematic review**

Ovid

Database(s): Embase 1988 to 2017 Week 31, EBM Reviews - Cochrane Central Register of Controlled Trials June 2017, EBM Reviews - Cochrane Database of Systematic Reviews 2005 to July 26, 2017, Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily and Ovid MEDLINE(R) 1946 to Present   
Search Strategy:

|  |  |  |
| --- | --- | --- |
| **#** | **Searches** | **Results** |
| 1 | exp bile duct cyst/ | 2475 |
| 2 | exp Bile Duct Diseases/ and exp Cysts/ | 8062 |
| 3 | (peribiliary or "peri-biliary" or periductal or "peri-ductal").ti,ab,hw,kw. | 3542 |
| 4 | (1 or 2) and 3 | 126 |
| 5 | ((peribiliary or "peri-biliary" or periductal or "peri-ductal") adj5 (cyst\* or polycyst\* or multicyst\*)).ti,ab,hw,kw. | 168 |
| 6 | 4 or 5 | 207 |
| 7 | remove duplicates from 6 | 126 |

Scopus

1. TITLE-ABS-KEY ( ( ( peribiliary  OR  "peri-biliary"  OR  periductal  OR  "peri-ductal" )  W/5  ( cyst\*  OR  polycyst\*  OR  multicyst\* ) ) )
2. PMID(0\*) OR PMID(1\*) OR PMID(2\*) OR PMID(3\*) OR PMID(4\*) OR PMID(5\*) OR PMID(6\*) OR PMID(7\*) OR PMID(8\*) OR PMID(9\*)
3. 1 and not 2

**Supplement 3**

**METHODS:**

**Data Sources and Search Strategies:** A comprehensive search of several databases from each database’s inception to August 2nd, 2017, any language was conducted. The databases included Ovid MEDLINE Epub Ahead of Print, Ovid Medline In-Process & Other Non-Indexed Citations, Ovid MEDLINE, Ovid EMBASE, Ovid Cochrane Central Register of Controlled Trials, Ovid Cochrane Database of Systematic Reviews, and Scopus. The search strategy was designed and conducted by an experienced librarian with input from the study’s principle investigator. Controlled vocabulary supplemented with keywords was used to search for peribiliary cysts. The actual strategy is provided in supplement II. In addition, the first 300 entries of Google Scholar were searched using the terms “peribiliary cysts” to look for studies not included in major databases. Reference lists were also manually reviewed for additional cases. The detailed search strategy is in Supplement 2.

**Inclusions criteria based on the following definitions:**

*Frequency of peribiliary cysts:* This is based on imaging, surgical or postmortem studies that evaluated peribiliary cysts.

*Diagnosis of peribiliary cysts:* This is done by one or more of the following imaging modalities: abdominal contrast-enhanced computed tomography (CECT), drip infusion cholangiographic computed tomography (DIC-CT), magnetic resonance imaging (MRI), magnetic resonance cholangio-pancreatography (MRCP); or examination of surgical or autopsy specimen.

*Clinical manifestations of peribiliary cysts:* This is based on case reports and case-series that reported sufficient clinical data

**Data Extraction and Assessment**

The data included year of publication; country of origin; publication language; publication format (full-text article, letter to the editors, image in medicine, abstract form); type of study (case report, case-series); age; gender; alcohol consumption; associated conditions or diseases [cirrhosis and etiology, portal hypertension, portal thrombosis, neoplasm, cholangitis, autosomal dominant polycystic kidney disease (ADPKD), autosomal dominant polycystic liver disease (ADPLD)]; clinical manifestations; liver function tests; tumor markers [carcino-embryologic antigen (CEA), carbohydrate antigen 19-9 (CA19-9)]; imaging modalities [abdominal contrast-enhanced computed tomography (CECT), drip infusion cholangiographic computed tomography (DIC-CT), abdominal magnetic resonance imaging (MRI), magnetic resonance cholangio-pancreatography (MRCP), endoscopic ultrasound (EUS), intraductal endoscopic ultrasound (ID-EUS), endoscopic retrograde cholangio-pancreatography (ERCP), percutaneous trans-hepatic cholangiography (PTC), endoscopic or intraoperative cholangioscopy]; accurate or inaccurate diagnosis of the disease; modalities allowing to make the diagnosis (imaging modalities, surgery or autopsy); number, size, and location of the cysts along the biliary tract; complications related to the disease; duration of follow-up after diagnosis and whether there was an increase in number and/or size of the cysts; and lastly final outcome, cause of death and postmortem exam.

**Assessment of methodological qualities of case reports and case series:**

Given that there are no available validated tools to assess the methodological quality of case reports and case series, we derived items from the Newcastle-Ottawa Scale (NOS) that were appropriate for this systematic review. We removed from the NOS the items that relate to comparability and adjustment (because the studies included were non-comparative). We retained for the purpose of methodological quality the items that focused on selection, representativeness of cases, and ascertainment of outcome and exposure This resulted in 5 criteria in the form of questions with a binary response (yes/no), whether the item was suggestive of bias or not. These questions are listed in Supplement 4. We considered the quality of the report good when all 5 criteria were fulfilled, moderate when 4 were fulfilled, and low when ≤3 were fulfilled.

**RESULTS:**

The flow diagram through the different phases of this systematic review is shown in Supplement 5. We identified 9 studies that assessed the frequency and 72 case reports and case series with sufficient clinical data [two of them were included to assess the frequency ([1](#_ENREF_1), [2](#_ENREF_2))].

**Studies characteristics:**

Seventy-twocase reports and case series from 10 countries published between 1984 and 2016 with sufficient clinical data were included. We identified 48 studies through the librarian search, 17 studies through manual search of reference lists ([3-19](#_ENREF_3)) and 7 studies through the Google Scholar database ([20-26](#_ENREF_20)). The publication language was Korean in 3 studies ([10](#_ENREF_10), [16](#_ENREF_16), [27](#_ENREF_27)), Japanese in 20 studies ([4-6](#_ENREF_4), [8](#_ENREF_8), [9](#_ENREF_9), [11-14](#_ENREF_11), [17](#_ENREF_17), [18](#_ENREF_18), [21-24](#_ENREF_21), [26](#_ENREF_26), [28-31](#_ENREF_28)), and English in the remaining studies. One publication was in an abstract form ([32](#_ENREF_32)), two were in form of letters to the editor ([33](#_ENREF_33), [34](#_ENREF_34)), four in form of image in medicine ([35-38](#_ENREF_35)) and all others were full-text manuscripts. There were 56 case reports and 16 case series that included 2 to 13 patients ([1-3](#_ENREF_1), [29-31](#_ENREF_29), [39-48](#_ENREF_39)). Supplement 7 demonstrates the number of reported cases from different countries. Most studies were reported from Japan [Japanese studies: 54/72 studies (75%) – Japanese cases 88/135 (65%)].

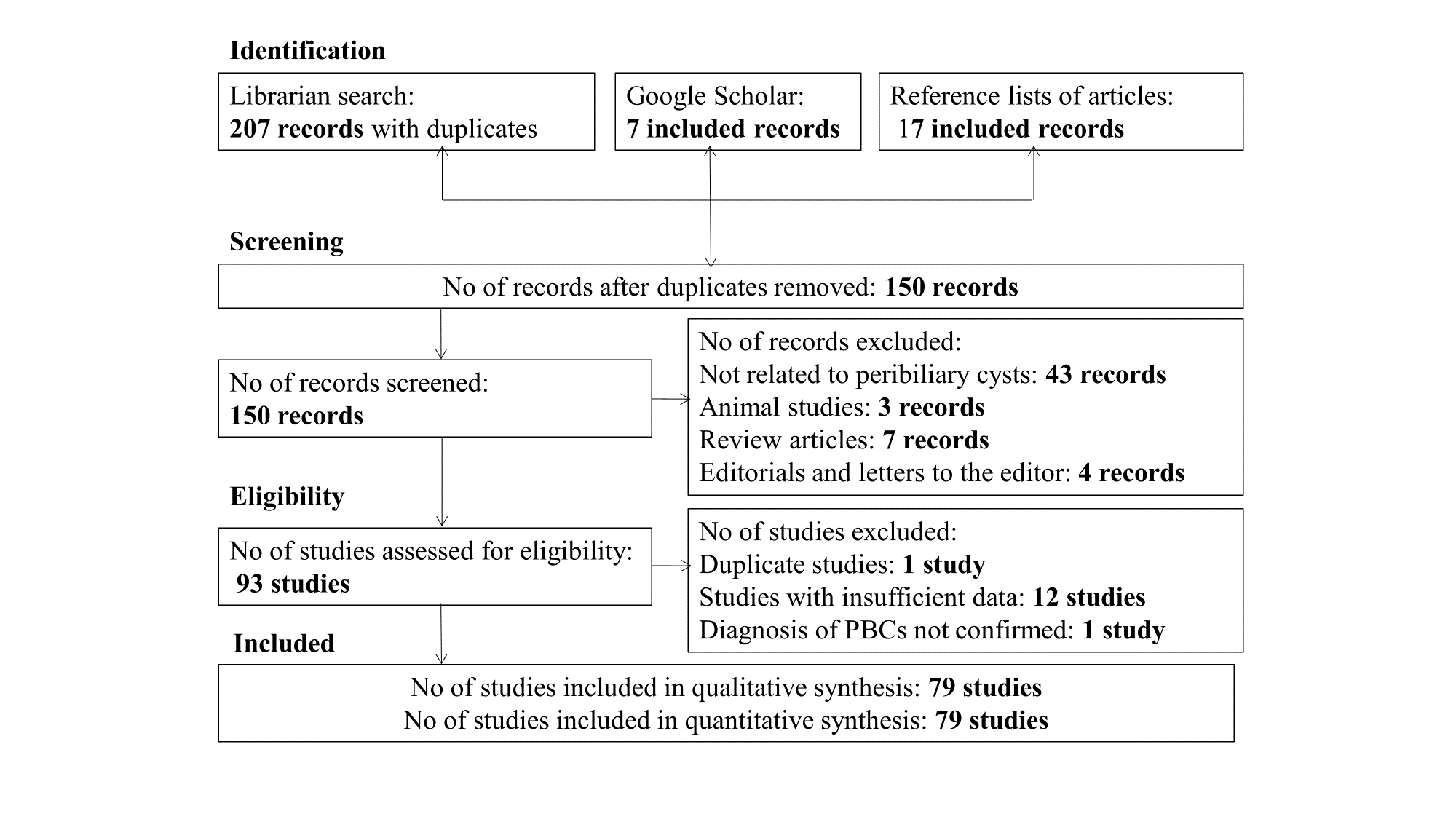
We excluded one duplicate study (Kim 2013) and 10 studies with insufficient clinical data some of them were duplicated (Qian 2003, Itai 2003, Brancatelli 2007, Matsubara 2012, Galia 2014, Matsubara 2014, Sato 2014, Katabathina 2016, Ronot 2017, Kozaka 2017). We excluded three patients with non-communicating cysts observed after Kasai’s operation for biliary atresia that were thought to be related to peribiliary glands without confirmation (Kawarasaki 1997), eight patients in the study of Colina et al. diagnosed as mucoceles of the cystic duct remnant in liver transplant patients (Colina 1999), and one patient with cyst in the wall of common bile duct that was thought to be related to a hematoma from the stump of the cystic artery during cholecystectomy (Scotiniotis 2001). References of excluded studies are found in supplement 8.

**Supplement 4**

|  |
| --- |
| **Tool for methodological qualities assessment of case reports and case series** |
| 1. Did the patient(s) represent the whole case(s) of the medical center? |
| 2. Was the diagnosis correctly made? |
| 3. Were other important diagnosis excluded? |
| 4. Were all important data cited in the report? |
| 5. Was the outcome correctly ascertained? |

**Supplement 5**

The flow diagram through the different phases of this systematic review



**Supplement 6. References of excluded studies**

|  |  |  |
| --- | --- | --- |
| **References** | **No of patients** | **Reason for exclusion** |
| **Duplicate study** | | |
| Kim H. Peribiliary cyst in healthy young patient. HPB 2013, 15 (Suppl. 2), 90A. | 1 patient | – |
| **Insufficient clinical data** | | |
| Qian Q, Li A, King BF, Kamath PS, Lager DJ, Huston J, et al. Clinical profile of autosomal dominant polycystic liver disease. Hepatology 2003;37(1):164–71. | – | study of autosomal dominant polycystic liver disease |
| Itai Y. Peribiliary cysts. Shoukaki Gazou (Journal of Gastroenterological Imaging). 2003; 5:35-40. | – | imaging study of peribiliary cysts |
| Brancatelli G, Federle MP, Ambrosini R et al. Cirrhosis: CT and MR imaging evaluation. European Journal of Radiology 2007;61: 57–69. | 1 patient | revue article of imaging for peribiliary cysts |
| Takashi Matsubara, Yasunori Sato, Motoko Sasaki, et al. Immunohistochemical characteristics and malignant progression of hepatic cystic neoplasms in comparison with pancreatic counterparts. Human Pathology 2012; 43, 2177–2186. | 10 patients | histological study without clinical details |
| Massimo Galia, Adele Taibbi, Daniele Marin et al. Focal lesions in cirrhotic liver: what else beyond hepatocellular carcinoma? Diagn Interv Radiol 2014; 20:222-228. | 1 patient | review article of imaging for peribiliary cysts |
| Matsubara T, Sato Y, Igarashi S et al. Alcohol-related injury to peribiliary glands is a cause of peribiliary cysts: based on analysis of clinical and autopsy cases. J Clin Gastroenterol 2014 Feb;48(2):153-9. | 60 patients |  |
| Yasunori Sato, Kenichi Harada, Motoko Sasaki, Yasuni Nakanuma? Cystic and micropapillary epithelial changes of peribiliary glands might represent a precursor lesion of biliary epithelial neoplasms. Virchows Arch 2014;464:157-163. | 49 patients | Histologic study of cystic change of peribiliary cysts (> 2 mm) |
| Katabathina VS, Flaherty EM, Dasyam AK et al. Biliary Diseases with Pancreatic Counterparts”: Cross-sectional Imaging Findings. RadioGraphics 2016; 36:374–392. | 3 patients | imaging study |
| Maxime Ronot, Marco Dioguardi Burgio, Yvonne Purcell, Romain Pommier, Giuseppe Brancatelli, Valerie Vilgrain. Focal lesions in cirrhosis: not always HCC. European Journal of Radiology 2017:93:157-168. | 1 patient | review article of focal lesions in cirrhosis |
| Kazuto Kozaka and Osamu Matsu. Pathology and Imaging of Peribiliary Cysts: Recent Progress. In: Y. Nakanuma (ed.), Pathology of the Bile Duct, Springer, Singapore, 2017, pp 113-120. | 2 patients | revue article of imaging for peribiliary cysts |
| Kawarasaki H, Itoh M, Mizuta K, Tanaka H, Makuuchi M. Further observations on cystic dilatation of the intrahepatic biliary system in biliary atresia after hepatic portoenterostomy: report on 10 cases. Tohoku J. Exp. Med.1997; 181: 175–83. | 3 patients | cysts after Kasai operation for biliary atresia |
| Colina F, Castellano VM, Gonzalez-Pinto I, et al. Hilar biliary cysts in hepatic transplantation. Report of three symptomatic cases and occurrence in resected liver grafts. Transpl Int 1998; 11:110–116. | 8 patients | mucoceles of cystic duct remnant in liver transplant patients |
| **Diagnosis of peribiliary cysts not confirmed** | | |
| Scotiniotis IA, Kochman ML. Intramural cyst of the bile duct demonstrated by cholangioscopy and intraductal US. Gastrointestinal Endoscopy 2001;54 (2): 260-262. | 1 patient | thought to be related to bleeding during cholecystectomy |
| **Total: 14 studies** |  |  |

**Supplement 7. Countries of reported studies of peribiliary cysts**

|  |  |  |  |
| --- | --- | --- | --- |
| **Countries** | **No of studies** | **No of reported cases** | **References** |
| Japan | 54 | 88 | Nakanuma 1984, Kasai 1984, Ueki 1987, Yoshinaga 1989, Terada 1992, Itai 1994, Terayama 1995, Akiyama 1997, Yuasa 1997, Terasaki 1997, Ishihara 1997, Fujioka 1997, Scolari 1997, Ahmadi 1997, Nagata 1998, Nishimura 1998, Takakura 2000, Miyake 2001, Motoo 2001, Okada 2001, Chiba 2002, Murai 2002, Cho 2003, Ohta 2003, Terada 2003, Kolodziejski 2004, Seguchi 2004, Shigeta 2005, Tsuchiya 2005, Yasutomo 2005, Yokomichi 2006, Otani 2006, Miura 2006, Ohnishi 2007, Kai 2008, Hamasu 2008, Ikenaga 2009, Tohma 2009, Nakanishi 2009, Fujiwara 2009, Sato 2009, Takahashi 2009, Nakayama 2010, Matsukura 2010, Terada 2010, Nakanishi 2011, Takuma 2012, Zen 212, Kozuki 2012, Teramoto 2015, Shibata 2015, Aoba 2015, Umemura 2016, Iijima 2016 |
| South Korea | 5 | 5 | Lee 1999, Park 2001, Kim 2009, Seo 2012, Kim 2013 |
| India | 2 | 14 | Gupta 1999, Johnson 2007 |
| Taiwan | 1 | 1 | Su 2013 |
| Australia | 1 | 2 | Pang 2010 |
| Spain | 1 | 5 | Colina 1998 |
| France | 2 | 2 | Da Ines 2011, Montoriol 2012 |
| UK | 1 | 1 | Fusai 2005 |
| USA | 4 | 15 | Herman 1990, Baron 1994, Stevens 1996, Lim 2016 |
| Canada | 1 | 2 | Wanless 1987 |
| **Total: 10 countries** | **72** | **135** |  |

**Supplement 8. Frequency of peribiliary cysts in imaging, surgical and postmortem studies**

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Author/Year** | **Country** | **Underlying condition** | **Diagnostic modality** | **No of patients** | **No of PBCs**  (%) | **Size of cysts**  mm |
| Terada/1990 ([49](#_ENREF_49)) | Japan | Postmortem | autopsy | 1000 | 202 (20.2%) | most microscopic  bile ducts compression in 16 |
| Kida/1992 ([50](#_ENREF_50)) | Japan | ADPLD | autopsy | 8 | 8 (100%) | all macroscopic |
| solitary nonparasitic cyst | 18 | 13 (72%) | 11microscopic-2macroscopic |
| normal liver | 23 | 8 (35%) | all microscopic |
| Itai/1995 ([51](#_ENREF_51)) | Japan | ADPKD & their relatives | CT or CECT | 64 | 22 (34%) | ˂10 mm - 30 mm in 1 patient |
| Hoshiba/1996 ([52](#_ENREF_52)) | Japan | liver cirrhosis | CT or CECT | 346 | 31 (9%) | ˂ 15 mm |
| no liver cirrhosis | 307 | 10 (3%) | ˂ 15 mm |
| Colina/1998 ([1](#_ENREF_1)) | Spain | liver transplant patients | resected graft/ autopsy | 493 | 5 (1%) | 5-20 mm |
| Gupta/1999 ([2](#_ENREF_2)) | India | ADPKD | CECT | 24 | 13 (54%) | 10 mm (some 10-30 mm) |
| Dranssart/2002 ([53](#_ENREF_53)) | France | ADPKD | MRCP | 90 | 59 (66%) | not reported |
| Matsubara 2014 ([54](#_ENREF_54)) | Japan | heavy alcohol use | autopsy | 202 | 29 (14%) | several mm to 20 mm |
| Goossens/2017 ([55](#_ENREF_55)) | Switzerland | liver transplant patients | resected graft | 71 | 17 (24%) | > 1 mm |
| **Total: 9 studies** | **5 countries** |  |  | **2646** | **417 (16%)** | **1-30 mm** |

ADPKD: autosomal dominant polycystic kidney disease – ADPLD: autosomal dominant polycystic liver disease – CECT: contrast enhanced computed tomography – CT: computed tomography – MRCP: magnetic resonance cholangiopancreatography – PBCs: peibiliary cysts.

**Supplement 9. Type of neoplasms associated with peribiliary cysts based on 135 cases**

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Type of neoplasms** | **No of neoplasm** | **Past history of neoplasm** | **Actual neoplasm** | **References** |
| Hepatocellular carcimoma | 16 | – | 16 | Nakanuma 1984, Wanless 1987, Terada 1992, Baron 1994, Terayama 1995, Ahmadi 1997, Seguchi 2004, Kai 2008, Tohma 2009, Zen 2012 |
| Intrahepatic cholangiocarcinoma1 | 1 | – | 1 | Otani 2006 |
| Extrahepatic cholangiocarcinoma | 1 | – | 1 | Miura 2006 |
| Intraductal papillary neoplasm of bile duct | 4 | – | 4 | Nakanishi 2009, Nakanishi 2011, Zen 2012 |
| Cancer of head of the pancreas | 1 | – | 1 | Fujiwara 2009 |
| Intraductal papillary neoplasm of pancreas | 1 | 1 | – | Sato 2009 |
| Lung cancer | 1 | – | 1 | Terada 2010 |
| Esophageal cancer | 1 |  | 1 | Takuma 2012 |
| Gastric cancer | 7 | 3 | 4 | Itai 1994, Akiyama 1997, Yuasa 1997, Ohta 2003, Seguchi 2004, Yasutomo 2005, Tohma 2009 |
| Colon cancer | 4 | 3 | 1 | Itai 1994, Nagata 1998, Ohta 2003, Aoba 2015 |
| Prostate cancer | 1 | 1 | – | Kolodziejski 2004 |
| Acute myeloid leukemia | 1 | – | 1 | Takahashi 2009 |
| **Total: 10 types of neoplasms** | **39 neoplasms**  **37 patients2** | **8** | **31** |  |

(1) diagnosis based on imaging and not confirmed histologically – (2) Two patients had 2 simultaneous neoplasms: hepatocellular carcinoma and early gastric cancer (Seguchi 2004), hepatocellular carcinoma and intraductal papillary neoplasm of the bile duct (Zen 2012).

**Supplement 10.** **Diagnostic modalities of peribiliary cysts based on 135 patients**

|  |  |  |
| --- | --- | --- |
|  | **No of patients** | **Correct diagnosis** |
| **Diagnosis without previous imaging modalities**  Diagnosis at surgery  Diagnosis at autopsy | **20**  9  11 | –  –  – |
| **Performance of one or more imaging modalities**  Correct diagnosis  Incorrect diagnosis  - Diagnosis at surgery  - Diagnosis at autopsy | **115**  69  46  37  9 | –  69/115 (60%)  46/115 (40%)  –  – |
| **Diagnostic accuracy of different imaging modalities**  CECT  DIC-CT  MRI  MRCP  Cholangioscopy1  IDUS | 105  19  20  39  12  4 | 51**/**105(48%)  15/19 (79%)  9/20 (45%)  25/39 (64%)  7/12 (58%)  3/4 (75%) |

(1) Cholangioscopy was performed during ERCP or intra-operatively and revealed submucosal lesions – CECT: contrast-enhanced computed tomography – DIC-CT: drip infusion cholangiographic computed tomography – EUS: endoscopic ultrasound – IDUS: intraductal ultrasound – MRCP: magnetic resonance cholangio-pancratography – MRI: magnetic resonance imaging.

**Supplement 11. Methodological quality assessment of 135 cases of peribiliary cysts**

|  |  |  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **First author/Year** | **No** | **Question 1** | | **Question 2** | | **Question 3** | | **Question 4** | | **Question 5** | | **Assessment** |
| **yes** | **no** | **yes** | **no** | **yes** | **no** | **yes** | **no** | **yes** | **no** |
| Nakanuma/1984([3](#_ENREF_3)) | 8 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 8 |
| Kasai/1984([4](#_ENREF_4)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Wanless/1987([39](#_ENREF_39)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Ueki/1987([5](#_ENREF_5)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Yoshinaga/1989([6](#_ENREF_6)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Herman/1990([56](#_ENREF_56)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Terada/1992([57](#_ENREF_57)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Baron/1994([40](#_ENREF_40)) | 12 | yes |  | yes |  | yes |  | yes |  | yes |  | good 12 |
| Itai/1994([42](#_ENREF_42)) | 4 | yes |  | yes |  | yes |  | yes |  | yes |  | good 4 |
| Terayama/1995([45](#_ENREF_45)) | 7 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 7 |
| Stevens/1996([58](#_ENREF_58)) | 1 | yes |  | yes |  | yes |  | yes | no | yes |  | moderate 1 |
| Akiyama/1997([7](#_ENREF_7)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Yuasa/1997([59](#_ENREF_59)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Terasaki/1997([44](#_ENREF_44)) | 2 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 2 |
| Ishihara/1997 ([28](#_ENREF_28)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Fujioka/1997([41](#_ENREF_41)) | 3 | yes |  | yes |  | yes |  | yes |  | yes |  | good 3 |
| Scolari/1997([19](#_ENREF_19)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Ahmadi/1997([60](#_ENREF_60)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Nagata/1998([29](#_ENREF_29)) | 3 | yes |  | yes |  | yes |  | yes |  | yes |  | good 3 |
| Nishimura/1998([8](#_ENREF_8)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Colina/1998([1](#_ENREF_1)) | 5 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 5 |
| Lee/1999([61](#_ENREF_61)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Gupta/1999([2](#_ENREF_2)) | 13 | yes |  | yes |  | yes |  |  | no |  | no | low 13 |
| Takakura/2000 ([9](#_ENREF_9)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Miyake/2001([35](#_ENREF_35)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Motoo/2001([62](#_ENREF_62)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Okada/2001([63](#_ENREF_63)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Park 2001 ([10](#_ENREF_10)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Chiba/2002([36](#_ENREF_36)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Murai/2002 ([11](#_ENREF_11)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Cho/2003 ([12](#_ENREF_12)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Ohta/2003 ([30](#_ENREF_30)) | 6 | yes |  | yes |  | yes |  | yes |  | yes |  | good 6 |
| Terada/2003([64](#_ENREF_64)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Kolodziejski/2004([20](#_ENREF_20)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Seguchi/2004([43](#_ENREF_43)) | 3 | yes |  | yes |  | yes |  | yes |  | yes |  | good 3 |
| Shigeta/2005 ([21](#_ENREF_21)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Tsuchiya/2005 ([13](#_ENREF_13)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Fusai/2005([65](#_ENREF_65)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Yasutomo/2005 ([14](#_ENREF_14)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Yokomichi/2006([66](#_ENREF_66)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Otani/2006 ([31](#_ENREF_31)) | 4 | yes |  | yes |  | yes |  | yes |  | yes |  | good 4 |
| Miura/2006([67](#_ENREF_67)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Johnson/2007([68](#_ENREF_68)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Ohnishi/2007([22](#_ENREF_22)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Kai/2008([69](#_ENREF_69)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Hamasu/2008 ([23](#_ENREF_23)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Ikenaga/2009([70](#_ENREF_70)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Nakanishi/2009([71](#_ENREF_71)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Tohma/2009([48](#_ENREF_48)) | 3 | yes |  | yes |  | yes |  | yes |  | yes |  | good 3 |
| Fujiwara/2009([34](#_ENREF_34)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Kim/2009([16](#_ENREF_16)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Sato/2009([72](#_ENREF_72)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Takahashi/2009([33](#_ENREF_33)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Pang/2010([46](#_ENREF_46)) | 2 | yes |  | yes |  | yes |  | yes |  | yes |  | good 2 |
| Nakayama/2010([37](#_ENREF_37)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Matsukura/2010([24](#_ENREF_24)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Terada/2010([73](#_ENREF_73)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Da Ines/2011([38](#_ENREF_38)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Nakanishi/2011([15](#_ENREF_15)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| Takuma/2012 ([18](#_ENREF_18)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Montoriol/2012([74](#_ENREF_74)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Zen/2012([47](#_ENREF_47)) | 1 | yes |  | yes |  | yes |  |  | no | yes |  | moderate 1 |
| 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Kozuki/2012 ([17](#_ENREF_17)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Seo/2012 ([27](#_ENREF_27)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Su/2013([25](#_ENREF_25)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Kim /2013([75](#_ENREF_75)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Teramoto /2015([32](#_ENREF_32)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Shibata/2015([76](#_ENREF_76)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Aoba/2015 ([26](#_ENREF_26)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Umemura/2016([77](#_ENREF_77)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Iijima/2016([78](#_ENREF_78)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| Lim/2016([79](#_ENREF_79)) | 1 | yes |  | yes |  | yes |  | yes |  | yes |  | good 1 |
| **Total: 72 studies** | 135 | 135 | 0 | 135 | 0 | 135 | 0 | 91 | 44 | 122 | 13 | good 91 (67%)  moderate 31 (23%)  low 13 (10%) |

**Supplement 12. Characteristic of peribiliary cysts according to their location along the biliary tract**

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Characteristics** | **Type I**  Intrahepatic  118 patients  Age 63 (4-88)1 | **Type II**  Extrahepatic  9 patients  Age 61 (43-79)1 | **Type III**  Intra- & extrahepatic  8 patients  Age 70.5 (53 -77)1 | **p value** |
| Males (%) | 89/105 (85%) | 2/9 (22%) | 6/8 (75%) | 0.001 |
| Japanese patients (%) | 74/118 (63%) | 5/9 (55%) | 6/8 (75%) | 0.784 |
| Associated conditions or diseases (%) | 104/118 (88%) | 6/9 (66%) | 7/8 (87.5%) | 0.168 |
| Cirrhosis (%) | 49/118 (41%) | 0/9 (0%) | 3/8 (37.5%) | 0.033 |
| Portal hypertension (%) | 37/101 (37%) | 0/9 (0%) | 3/7 (43%) | 0.046 |
| Hepatic, biliary or pancreatic neoplasms (%) | 23/118 (19.5%) | 0//9 (0%) | 0/8 (0%) | 0.197 |
| Incorrect diagnosis by imaging modalities (%) | 38/104 (36%) | 5/6 (83%) | 4/5 (80%) | 0.012 |
| Solitary cysts (%) | 4/118 (3.4%) | 9/9 (100%) | 0/8 (0%) | 0.001 |
| Dilated biliary tract (%) | 38/111 (34%) | 4/9 (44%) | 5/6 (83%) | 0.057 |
| Obstructive jaundice (%) | 12/111 (11%) | 4/9 (44%) | 2/6 (33%) | 0.012 |
| Cholangitis (%) | 12/89 (13%) | 1/9 (11%) | 1/6 (17%) | 1.000 |
| Surgical interventions (%) | 49/118 (41.5%) | 8/9 (89%) | 4/8 (50%) | 0.019 |
| Therapeutic misadventures (%) | 18/49 (37%) | 1/8 (12.5) | 1/4 (25%) | 0.517 |
| Mortality rate (%) | 25/105 (24%) | 1/9 (11%) | 3/8 (37.5%) | 0.461 |

(1) Median age and range.

**Supplement 13. Comparison between multiple versus solitary cysts**

|  |  |  |  |
| --- | --- | --- | --- |
| **Characteristics** | **Multiple cysts**  122 patients  Age 64 (4-88)1 | **Solitary cysts**  13 patients  Age 61 (9-81)1 | **p value** |
| Males (%) | 94/109 (86%) | 4/13 (31%) | 0.0027 |
| Japanese patients (%) | 78/122 (64%) | 7/13 (54%) | 0.474 |
| Associated conditions or diseases (%) | 109/122 (89%) | 6/13 (46%) | 0.0001 |
| Cirrhosis (%) | 51/122 (42%) | 1/13 (8%) | 0.0166 |
| Portal hypertension (%) | 40/106 (38%) | 0/11 (0%) | 0.0152 |
| Hepatic, biliary or pancreatic neoplasms (%) | 23/122 (19%) | 0/13 (0%) | 0.2180 |
| Incorrect diagnosis by imaging modalities (%) | 40/107 (37%) | 7/8 (87%) | 0.0078 |
| Dilated biliary tract (%) | 42/113 (37%) | 5/13 (38%) | 0.9272 |
| Obstructive jaundice (%) | 14/113 (12%) | 4/13 (31%) | 0.2306 |
| Cholangitis (%) | 12/92 (13%) | 1/12 (8%) | 1.0000 |
| Surgical interventions (%) | 49/122 (40%) | 12/13 (92%) | 0.2097 |
| Therapeutic misadventures (%) | 19/49 (39%) | 1/ 12 (8%) | 0.0826 |
| Mortality rate (%) | 28/109 (26%) | 1/13 (8%) | 0.1878 |

(1) Median age and range.

**REFERENCES OF INCLUDED STUDIES**

1. Colina F, Castellano VM, Gonzalez-Pinto I, Garcia I, Novo O, Garcia-Hidalgo E, Garcia-Munoz H, et al. Hilar biliary cysts in hepatic transplantation. Report of three symptomatic cases and occurrence in resected liver grafts. Transplant International 1998;11:110-116.

2. Gupta S, Seith A, Dhiman RK, Chawla YK, Sud K, Kohli HS, Sakhuja V, et al. CT of liver cysts in patients with autosomal dominant polycystic kidney disease. Acta Radiologica 1999;40:444-448.

3. Nakanuma Y, Kurumaya H, Ohta G. Multiple cysts in the hepatic hilum and their pathogenesis. Virchows Archiv A 1984;404:341-350.

4. Kasai Y, Kakita A, Takahashi T, al. e. Intramural cyst of common bile duct combined with polycystic disease. Fukubu Gazou Shindan (Diagn Image Abdom). 1984;4:430-132.

5. Ueki Y, Mori T, Tominaga Y, Kusumoto M, Nakata K, Nagataki S. A case of simple cyst derived from the common bile duct; undiscriminated from malignant lesion. Nihon Naika Gakkai zasshi. The Journal of the Japanese Society of Internal Medicine 1987;76:1889.

6. Yoshinaga T, Sugiyama T, Kimura M, al. e. A case of intramural cyst of the bile duct at the junction of the cystic and common bile duct. Shoukakinaishikyou no Shinpo (Prog Dig Endosc) 1989;35:381-383.

7. Akiyama H, Nimura Y, Kondo S, Kamiya J, Nagino M, Miyachi M, Kanai M. Multiple intramural cysts of the bile duct. Gastrointestinal endoscopy 1997;45:310-312.

8. Nishimura T, Suda M, Ueno M, Iwanaga M. A case of intramural bile duct cysts of right hepatic duct. Geka (Surgery) 1998;60:841-844.

9. Takakura N, Shima Y, Hamada M, al. e. A resected case of hepatic peribiliary cysts. . Kan Tan Sui 2000;40:313-316.

10. Park JS, Lee DH, Lim JW, Ko YT, Lee SM, Kim YH. Peribiliary Cysts with Intrahepatic Bile Duct Obstruction: A Case Report. Journal of the Korean Radiological Society 2001;45:615-619.

11. Murai S, Matsubara K, Fujita K, al. e. A case report of hepatobiliary cyst adenoma in men. Tan to Sui (Journal of Biliary Tract & Pancreas) 2002;23:77-80.

12. Cho A, Ochiai T, Tohma T, al. e. A case of multiple hepatic peribiliary cysts difficult to differentiate from cholangiocellular carcinoma. Syujyutu (Operation) 2003;5:1281-1284

13. Tsuchiya S, Tsuyuguchi T, Sakai Y, Kobayashi A, Fukuda Y, H. S. A case of peribiliary cysts diagnosed by intraductal ultrasonography. Tando (Journal of Japan Biliary Association) 2005;18:639-644.

14. Yasutomo N, Kawabata M, T. G. Hepatic peribiliary cysts diagnosed after surgery for gastric carcinoma and strangulated ileus. Rinsho Geka (J Clin Surg) 2005;60:903-906.

15. Nakanishi Y, Nakanuma Y, Ohara M, Iwao T, Kimura N, Ishidate T, Kijima H. Intraductal papillary neoplasm arising from peribiliary glands connecting with the inferior branch of the bile duct of the anterior segment of the liver. Pathology international 2011;61:773-777.

16. Kim PJ, Kang DH, Jeong WJ, Kim ID, Eum JS, Choi CW, Song GA. A case of peribiliary cysts. Korean Journal of Gastrointestinal Endoscopy 2009;38:368-370.

17. Kozuki A, Shima Y, Sumiyoshi T, J. I. A resected case of hepatic peribiliary cysts difficult to distinguish from intrahepatic cholangiocarcinoma. Tan to Sui (Journal of Biliary Tract & Pancreas) 2012;26:604-609.

18. Takuma K, Kamizawa T, Hara S, Kuruma S, Tabata T, K. C. Images of hepatic peribiliary cysts. Tando (Journal of Japan Biliary Association) 2012;26:136-139.

19. Scolari F, Valzorio B, Carli O, Vizzardi V, Costantino E, Grazioli L, Bondioni M, et al. Oral-facial-digital syndrome type I: an unusual cause of hereditary cystic kidney disease. Nephrology, dialysis, transplantation: official publication of the European Dialysis and Transplant Association-European Renal Association 1997;12:1247-1250.

20. Kolodziejski TR, Safadi BY, Nakanuma Y, Milkes DE, Soetikno RM. Bile duct cysts in a patient with autosomal dominant polycystic kidney disease. Gastrointestinal endoscopy 2004;59:140-142.

21. Shigeta M, Sudo M, Orita M, Enoki T, Noshima S, Hamano K. A Case of Hilar Bile Duct Stenosis Caused by Peribiliary Cysts. Nihon Rinsho Geka Gakkai Zasshi (Journal of Japan Surgical Association) 2005;66:1166-1169.

22. Ohnishi K, Ishida M, Iida A, Katayama K, Yamaguchi A, Y I. A case of hepatic peribiliary cysts with difficult pre-operative diagnosis. Nihon Rinsho Geka Gakkai Zasshi (Journal of Japan Surgical Association) 2007;68:1775-1780.

23. Hamasu S, Horii S, Sato B, Y. U. A case of hepatic peribiliary cysts resembling intrahepatic cholangiocarcinoma. . Rinsho Geka (J Clin Surg) 2008;69:2965-2969.

24. Matsukura M, Chida T, Kageyama F, Ohta K, Matsushita N, Shimoyama M, al. e. A case of intrahepatic choledochal stone and peribiliary cysts with bile duct stricture. Journal of Hamamatsu Medical Center 2010;4.

25. Su WL, Chang WT, Wang SN, Chuang SC, Kuo KK, Lee KT. Solitary peribiliary cyst mimicking cholangiocarcinoma. Formosan Journal of Surgery 2013;46:61-64.

26. Aoba T, Kato T, Hiramatsu K. A resected case of hepatic peribiliary cysts undedicated preoperative diagnosis. Tando (Journal of Japan Biliary Association) 2015;29:266-270.

27. Seo JY, Oh TH, Jeon TJ, Seo DD, Shin WC, Choi WC. [A case of hepatic peribiliary cysts in a patient with alcoholic liver cirrhosis]. Korean Journal of Gastroenterology/Taehan Sohwagi Hakhoe Chi 2012;60:119-122.

28. Ishihara A, Hukohe Y, Koizumi K, Murakami M, Kusano M. A case of a small hilar cyst derived from the peribiliary gland with compression of the intrahepatic bile duct. Japanese Journal of Gastroenterological Surgery 1997;30:1937-1941.

29. Nagata T, Imashuku Y, Maeda K, Yamauchi H. Hepatic peribiliary cyst on MRCP. [Japanese]. Japanese Journal of Clinical Radiology 1998;43:587-592.

30. Ohta H, Baba Y, Ishikawa T, Yosida T, Kamimura T, Tsubono T. A clinical evaluation of hepatic peribiliary cysts. [Japanese]. Acta Hepatologica Japonica 2003;44:58-65.

31. Otani K, Ueki T, Kawamoto K, Noma E, Shimizu A, Fujimura N, Sakaguchi S, et al. [Clinical course of 4 cases of hepatic peribiliary cysts and a summary of the 38 clinical cases reported in Japan]. Nippon Shokakibyo Gakkai Zasshi - Japanese Journal of Gastroenterology 2006;103:961-968.

32. Teramoto A, Nakamatsu G, Shimizu S, Fukuhara T, Kimura N, Kondo A, Iraha A, et al. A case of peribiliary cyst infection due to hepatolithiasis. American Journal of Gastroenterology 2015;110:S51-S52.

33. Takahashi H, Miwa S, Nomoto K, Hayashi S, Tsuneyama K, Takano Y. Peribiliary cyst of common bile duct: possible cause of refractory jaundice. Pathology International 2009;59:692-693.

34. Fujiwara H, Gobara H, Mimura H, Hiraki T, Iguchi T, Kanazawa S. Sclerotherapy for Peribiliary Cysts Accompanied by Biliary Stenosis. Journal of Vascular and Interventional Radiology 2009;20:1644-1645.

35. Miyake H, Yuasa N, Kamiya J, Nagino M, Uesaka K, Oda K, Sano T, et al. Images in focus. Peribiliary cysts both in the cystic duct and in the intrahepatic biliary tract. Endoscopy 2001;33:643.

36. Chiba M, Obata H. Cholangiography in a patient with hilar peribiliary cysts. Journal of Hepatology 2002;37:288.

37. Nakayama S. Development of multiple peribiliary cysts following acute alcoholic hepatitis. Indian Journal of Gastroenterology 2010;29:129.

38. Da Ines D, Essamet W, Montoriol PF. Peribiliary cysts. Hepatology 2011;54:2271-2272.

39. Wanless IR, Zahradnik J, Heathcote EJ. Hepatic cysts of periductal gland origin presenting as obstructive jaundice. Gastroenterology 1987;93:894-898.

40. Baron RL, Campbell WL, Dodd GD, 3rd. Peribiliary cysts associated with severe liver disease: imaging-pathologic correlation. AJR American Journal of Roentgenology 1994;162:631-636.

41. Fujioka Y, Kawamura N, Tanaka S, Fujita M, Suzuki H, Nagashima K. Multiple hilar cysts of the liver in patients with alcoholic cirrhosis: report of three cases. Journal of Gastroenterology & Hepatology 1997;12:137-143.

42. Itai Y, Ebihara R, Tohno E, Tsunoda HS, Kurosaki Y, Saida Y, Doy M. Hepatic peribiliary cysts: multiple tiny cysts within the larger portal tract, hepatic hilum, or both. Radiology 1994;191:107-110.

43. Seguchi T, Akiyama Y, Itoh H, Tanaka H, Naganuma S, Nagaike K, Uchiyama S, et al. Multiple hepatic peribiliary cysts with cirrhosis. Journal of Gastroenterology 2004;39:384-390.

44. Terasaki S, Nakanuma Y, Unoura M, Kaneko S, Kobayashi K. Involvement of peribiliary glands in primary sclerosing cholangitis: a histopathologic study. Internal Medicine 1997;36:766-770.

45. Terayama N, Matsui O, Hoshiba K, Kadoya M, Yoshikawa J, Gabata T, Takashima T, et al. Peribiliary cysts in liver cirrhosis: US, CT, and MR findings. Journal of Computer Assisted Tomography 1995;19:419-423.

46. Pang T, Kuo S, Hugh TJ, Davidson T, Eckstein RP, Bell C, Samra JS. The role of peribiliary cysts in biliary obstruction. ANZ Journal of Surgery 2010;80:699-702.

47. Zen Y, Amarapurkar AD, Portmann BC. Intraductal tubulopapillary neoplasm of the bile duct: potential origin from peribiliary cysts. Human Pathology 2012;43:440-445.

48. Tohma T, Miura F, Cho A, Okazumi S, Asano T. Usefulness of computed tomography during cholangiography for the diagnosis of multiple hepatic peribiliary cysts: a report of three cases with chronic liver disease. Journal of Hepato-Biliary-Pancreatic Surgery 2009;16:372-375.

49. Terada T, Nakanuma Y. Pathological observations of intrahepatic peribiliary glands in 1,000 consecutive autopsy livers. III. Survey of necroinflammation and cystic dilatation. Hepatology 1990;12:1229-1233.

50. Kida T, Nakanuma Y, Terada T. Cystic dilatation of peribiliary glands in livers with adult polycystic disease and livers with solitary nonparasitic cysts: an autopsy study. Hepatology 1992;16:334-340.

51. Itai Y, Ebihara R, Eguchi N, Saida Y, Kurosaki Y, Minami M, Araki T. Hepatobiliary cysts in patients with autosomal dominant polycystic kidney disease: prevalence and CT findings. AJR American Journal of Roentgenology 1995;164:339-342.

52. Hoshiba K, Matsui O, Kadoya M, Yoshikawa J, Gabata T, Terayama N, Takashima T. Peribiliary cysts in cirrhotic liver: observation on computed tomography. Abdominal Imaging 1996;21:228-232.

53. Dranssart M, Cognet F, Mousson C, Cercueil JP, Rifle G, Krause D. MR cholangiography in the evaluation of hepatic and biliary abnormalities in autosomal dominant polycystic kidney disease: study of 93 patients. Journal of Computer Assisted Tomography 2002;26:237-242.

54. Matsubara T, Sato Y, Igarashi S, Matsui O, Gabata T, Nakanuma Y. Alcohol-related injury to peribiliary glands is a cause of peribiliary cysts: based on analysis of clinical and autopsy cases. Journal of Clinical Gastroenterology 2014;48:153-159.

55. Goossens N, Breguet R, De Vito C, Terraz S, Lin-Marq N, Giostra E, Rubbia-Brandt L, et al. Peribiliary Gland Dilatation in Cirrhosis: Relationship with Liver Failure and Stem Cell/Proliferation Markers. Digestive diseases and sciences 2017;62:699-707.

56. Herman TE, Siegel MJ. Central dot sign on CT of liver cysts. Journal of Computer Assisted Tomography 1990;14:1019-1021.

57. Terada T, Minato H, Nakanuma Y, Shinozaki K, Kobayashi S, Matsui O. Ultrasound visualization of hepatic peribiliary cysts: a comparison with morphology. American Journal of Gastroenterology 1992;87:1499-1502.

58. Stevens W, Harford W, Lee E. Obstructive jaundice due to multiple hepatic peribiliary cysts. American Journal of Gastroenterology 1996;91:155-157.

59. Yuasa N, Nimura Y, Hayakawa N, Kamiya J, Maeda S, Kondo S, Nagasaka T. Multiple hepatic cysts along the intrahepatic bile duct--case report. Hepato-Gastroenterology 1997;44:1262-1266.

60. Ahmadi T, Itai Y, Minami M. Central dot sign in entities other than Caroli disease. Radiation Medicine 1997;15:381-384.

61. Lee JM, Jung SE, Lee KY, Kim SN, Sun HS. A large peribiliary cyst in the extrahepatic bile duct. AJR American Journal of Roentgenology 1999;173:621-623.

62. Motoo Y, Yamaguchi Y, Watanabe H, Okai T, Sawabu N. Hepatic peribiliary cysts diagnosed by magnetic resonance cholangiography. Journal of Gastroenterology 2001;36:271-275.

63. Okada S, Kojima Y, Morozumi A, Ainota T, Miyazaki Y, Akahane Y. Multiple hepatic peribiliary cysts discovered incidentally at a medical examination. Journal of Gastroenterology & Hepatology 2001;16:1167-1169.

64. Terada T, Matsushita H, Tashiro J, Sairenji T, Eriguchi M, Osada I. Cholesterol hepatolithiasis with peribiliary cysts. Pathology International 2003;53:716-720.

65. Fusai G, Tucker O, Nik Sulaiman NM, Karani J, Rela M, Portmann B. Peribiliary cysts can mimic Caroli's disease: a case report. International Journal of Surgical Pathology 2005;13:379-382.

66. Yokomichi H, Tsuji K, Hayashi Y, Kaneko M, Nakadoi K, Ishida Y, Kuwabara T, et al. A case of multiple hepatic peribiliary cysts which contributed to the obstructive jaundice and led to liver failure at the young man with von Recklinghausen's disease. Hepatology Research 2006;35:222-227.

67. Miura F, Takada T, Amano H, Yoshida M, Isaka T, Toyota N, Wada K, et al. A case of peribiliary cysts accompanying bile duct carcinoma. World Journal of Gastroenterology 2006;12:4596-4598.

68. Johnson MA, Ravichandran P, Surendran R. Solitary extra-hepatic hilar peribiliary cyst presenting with obstructive jaundice: a case report. Acta Chirurgica Belgica 2007;107:716-719.

69. Kai K, Eguchi Y, Kumagai T, Sugita Y, Tokunaga O. An autopsy case of obstructive jaundice due to hepatic multiple peribiliary cysts accompanying hepatolithiasis. Hepatology Research 2008;38:211-216.

70. Ikenaga N, Chijiiwa K, Otani K, Ohuchida J, Uchiyama S. A case of peribiliary cyst presenting with obstructive jaundice. Journal of Gastrointestinal Surgery 2009;13:174-176.

71. Nakanishi Y, Zen Y, Hirano S, Tanaka E, Takahashi O, Yonemori A, Doumen H, et al. Intraductal oncocytic papillary neoplasm of the bile duct: the first case of peribiliary gland origin. Journal of Hepato-Biliary-Pancreatic Surgery 2009;16:869-873.

72. Sato Y, Mukai M, Sasaki M, Kitao A, Yoneda N, Kobayashi D, Imamura Y, et al. Intraductal papillary-mucinous neoplasm of the pancreas associated with polycystic liver and kidney disease. Pathology International 2009;59:201-204.

73. Terada T, Moriki T. Monolobar ductal plate malformation disease of the liver. Pathology International 2010;60:407-412.

74. Montoriol PF, Poincloux L, Petitcolin V, Da Ines D. Peribiliary cysts mistaken for a biliary dilatation in a cirrhosis patient. Clinics & Research in Hepatology & Gastroenterology 2012;36:e93-95.

75. Kim HJ, Kim CY, Hur YH, Kim JC, Cho CK, Kim HJ. Peribiliary cysts developed in normal underlying liver: report of a case. Korean Journal of Hepatobiliarypancreatic Surgery 2013;17:131-134.

76. Shibata M, Hiura M, Senju M, Matsuhashi T, Abe S, Morita C, Hayashida K, et al. Hepatic peribiliary cysts with rapidly progressive refractory obstructive jaundice and esophageal varices. Internal Medicine 2015;54:389-393.

77. Umemura A, Suto T, Sasaki A, Nitta H, Nakamura S, Endo F, Harada K, et al. Pure Laparoscopic Left Hemihepatectomy for Hepatic Peribiliary Cysts with Biliary Intraepithelial Neoplasia. Case Reports in Surgery 2016;2016:7236427.

78. Iijima T, Hoshino J, Mise K, Sumida K, Suwabe T, Hayami N, Ueno T, et al. Daughter and mother with orofaciodigital syndrome type 1 and glomerulocystic kidney disease. Human Pathology 2016;55:24-29.

79. Lim J, Nissen NN, McPhaul C, Annamalai A, Klein AS, Sundaram V. Peribiliary hepatic cysts presenting as hilar cholangiocarcinoma in a patient with end-stage liver disease. Journal of Surgical Case Reports 2016;8:10.