



Case report

Gallbladder cancer spreading into the aberrant cystic duct: First literature report

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ABSTRACT

Introduction and importance: Although variations from the standard anatomy of the extrahepatic bile ducts are common, duplication of the cystic duct draining a single gallbladder is an extremely rare variant. We herein describe the first report of gallbladder cancer spreading into the aberrant cystic duct.

Case presentation: A 60-year-old female presented with upper abdominal pain, and she was diagnosed with gallbladder cancer. Intraoperatively, she was found to have a duplicated cystic duct draining a single gallbladder, and her cancer had spread into the aberrant cystic duct entering the anterior right hepatic duct. Right hepatectomy with extrahepatic bile duct resection was performed to achieve R0 resection.

Clinical discussion: In the English literature, 28 cases of duplicated cystic duct draining a single gallbladder have been reported. However, no cases of gallbladder cancer have been described in these previous reports.

Conclusion: We report the first case of gallbladder cancer spreading into the aberrant cystic duct. To perform an oncologically adequate operation, exact assessment of the biliary tree is essential not only preoperatively but also intraoperatively.

1. Introduction

Anomalies and variations in the anatomy of the biliary tract have been reported to occur in up to 47% of the population based on operative, cholangiographic, and autopsy studies [1]. Unlike the more common variant, in which two cystic ducts drain two distinct gallbladders, one of the extremely rare abnormalities of the cystic duct is a duplicated cystic duct draining a single gallbladder, which is usually discovered intraoperatively. Herein, we report the first case of gallbladder cancer spreading into an aberrant cystic duct diagnosed intraoperatively as a duplicated cystic duct draining a single gallbladder. This report is in line with the SCARE criteria [2].

2. Case presentation

A 60-year-old female presented to the emergency department with the chief complaint of upper abdominal pain. Her history of illness included diabetes mellitus, hypertension and dyslipidemia, which were medically treated. Physical examination revealed upper abdominal

tenderness without peritoneal signs and a negative clinical Murphy's sign. Jaundice was observed in the skin and conjunctiva. Laboratory studies showed an elevated white blood cell count (WBC: 11,800/ μ L), increased aspartate aminotransferase (AST: 1321 IU/L), alanine aminotransferase (ALT: 1165 IU/L), and γ -glutamyl transpeptidase (γ -GTP: 233 IU/L) levels, and high levels of the tumour marker carbohydrate antigen 19-9 (CA19-9: 129.7 U/mL). Ultrasonography revealed a gallbladder tumour spreading from the gallbladder neck to the body. She was admitted to our hospital for further examination.

Abdominal ultrasonography, computed tomography (CT) and magnetic resonance imaging (MRI) revealed a 3 cm gallbladder tumour with suspicion of invasion to the liver bed (Fig. 1a, b, c, d). This gallbladder tumour was enhanced on CT and showed a high signal on diffusion-weighted magnetic resonance imaging (DW-MRI) (Fig. 1e). Endoscopic retrograde cholangiography (ERC) demonstrated that the aberrant cystic duct drained to the anterior right hepatic duct (Fig. 1f), and bile juice cytology via a nasobiliary drainage tube resulted in false positives.

Thus, the patient was diagnosed with gallbladder cancer with a

Abbreviations: ERCP, endoscopic retrograde cholangiopancreatography; IFSA, intraoperative frozen section analysis; BiIIN, intraepithelial neoplasm.

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variation in the cystic duct entering the anterior right hepatic duct. We planned cholecystectomy plus liver bed resection and lymph node dissection in the hepatoduodenal ligament, and extrahepatic bile duct resection or liver resection was considered to be necessary depending on the level of tumour invasion. On laparotomy, we first found that the main cystic duct entered the common bile duct (Fig. 2a). At this point, we identified duplicated cystic ducts draining a single gallbladder. To assess the necessity of extrahepatic bile duct resection, we performed intraoperative frozen section analysis (IFSA). The 1st IFSA revealed a biliary intraepithelial neoplasm (BilIN) in the cut end of the main cystic duct. Then, we performed 2nd IFSA of the bifurcation of the common bile duct (CBD) and main cystic duct and found another BilIN. Both IFSA of the CBD and confluence of the right and left hepatic ducts also revealed BilINs, so we determined that extrahepatic bile duct resection was needed. Subsequently, we transitioned to liver bed resection and began liver parenchymal transection using the Cavitron ultrasonic surgical aspirator (CUSA). We exposed the aberrant cystic duct draining to the anterior right hepatic duct during liver parenchymal transection (Fig. 2b). IFSA of the aberrant cystic duct was performed, which showed adenocarcinoma. Therefore, we diagnosed the patient as having gallbladder cancer spreading to the aberrant cystic duct that drained into the anterior right hepatic duct, and we decided to perform right hepatectomy with extrahepatic bile duct resection to achieve R0 resection. After dissecting the right hepatic artery and right portal vein at the hepatic hilum, we completed liver parenchymal transection followed by dissection of the left hepatic duct and right hepatic vein. Right hepatectomy with extrahepatic bile duct resection and hepaticojejunostomy were performed. The operative time and bleeding volume were 653 minutes and 453 mL, respectively.

Histopathological examination of the gallbladder revealed papillary adenocarcinoma spreading from the gallbladder neck to the aberrant cystic duct (pT1b, pN0, pM0 and pStage IB) and widespread BilINs in the extrahepatic bile duct (Fig. 3).

Biliary leakage, pleural effusion and intra-abdominal abscess

occurred postoperatively; percutaneous drainage was performed, and the patient was discharged from the hospital on postoperative day 102. She remained free of recurrence for 33 months after the operation.

3. Discussion

Although anatomical variations in the cystic duct are quite common, a duplicate cystic duct draining a single gallbladder is an extremely rare anomaly of the biliary tract, with 29 cases having been reported in the literature worldwide, including our case [1,3–28]. In addition, no report of gallbladder cancer spreading into the aberrant cystic duct was found, so our present case is the first case report in the global literature.

Caster and Flannery classified this anomaly into 3 types, as follows [29]: The first category is the “H” type, in which the aberrant cystic duct enters separately into the right, left or common hepatic duct. This is the most frequent type of variation in the published literature, and our case was also of this type. The second category is the “Y” type, in which the 2 cystic ducts meet to form a common channel that enters the common hepatic duct. The third category is the “trabecular” type, in which the aberrant cystic duct directly enters the substance of the liver. Table 1 shows a review of the cases of duplicated cystic ducts draining a single gallbladder in the English literature. Twenty-three of these 29 patients (79%) were female, and the “H” type variation was the most common type.

A previous multi-institutional study defined the clinicopathological characteristics and role of surgery in patients with primary cystic duct cancer [30]. According to that study, cystic duct cancer is characterized by locally advanced disease with aggressive histopathological characteristics at the time of surgery; thus, extensive resection is often required during treatment. The classification of pN and pM were independent prognostic factors associated with both overall and disease-specific survival, indicating that radical resection provides potential benefits for patients with pN0pM0 disease, whereas pN1/2 and/or pM1 status appear to have strong adverse effects on survival. In our case, it was so

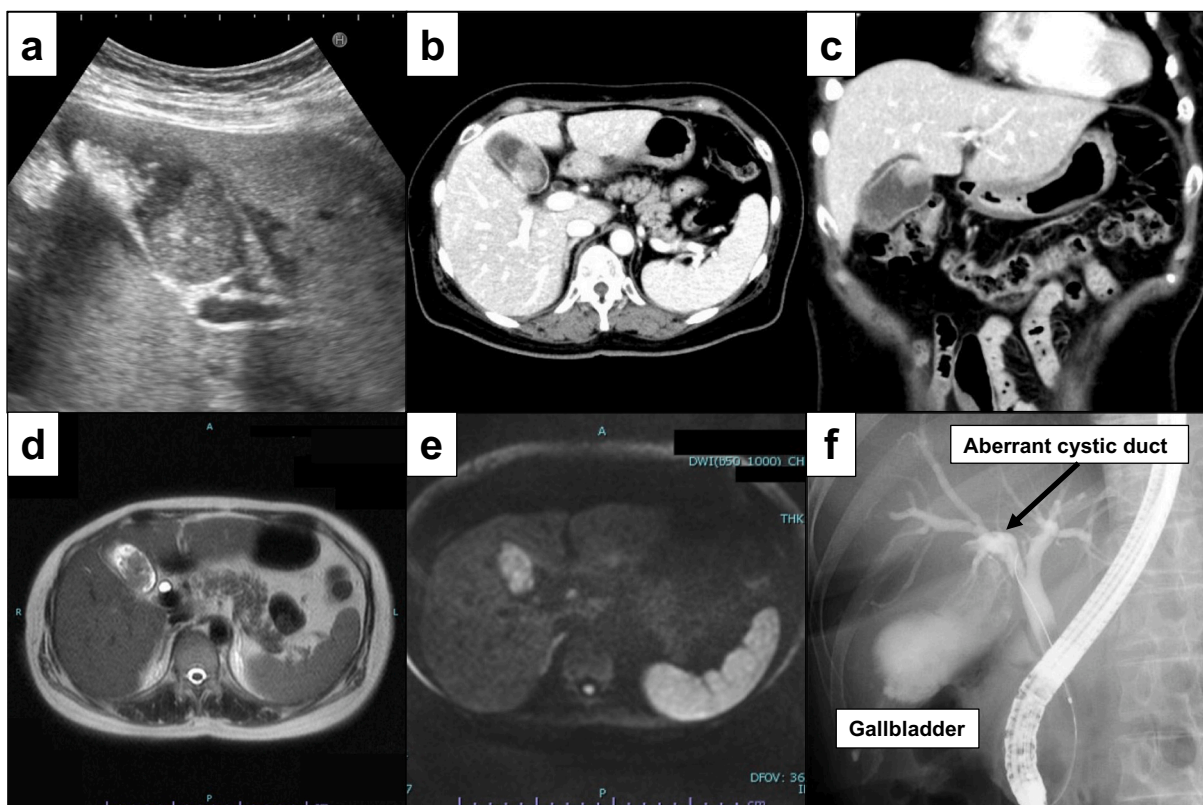


Fig. 1. a: US, b: axial CT, c: coronal CT, d: MRI, e: DW-MRI, f: ERC.

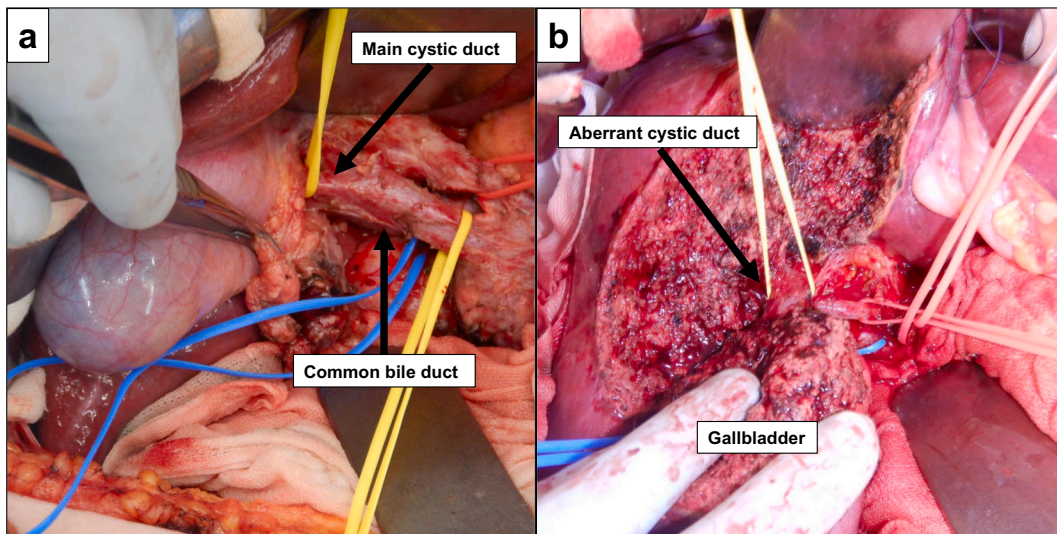


Fig. 2. a: Main cystic duct entering the common bile duct, b: aberrant cystic duct draining to the anterior right hepatic duct.

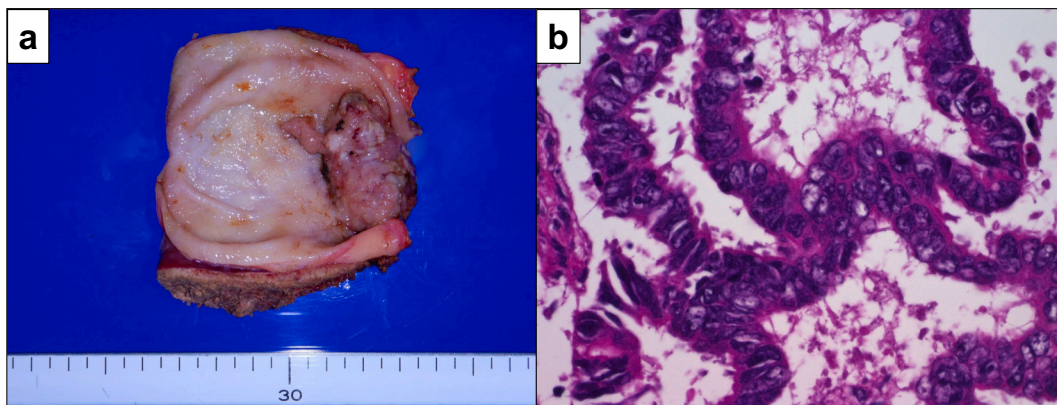


Fig. 3. a: Resected specimen, b: histopathological examination showing papillary adenocarcinoma spreading from the gallbladder neck to the aberrant cystic duct.

difficult to identify whether the patient's cancer invaded the anterior right hepatic duct that exposure of the aberrant cystic duct intrahepatically during liver parenchymal dissection and IFSA were essential to determine the appropriate surgical procedure. Finally, we completed R0 resection by right hepatectomy with extrahepatic bile duct resection, and histopathological examination revealed stage IB (pN0, pM0) gallbladder cancer spreading into the aberrant cystic duct.

4. Conclusion

We report the first case of gallbladder cancer spreading into the aberrant cystic duct. To perform an oncologically adequate operation, exact assessment of the biliary tree is essential not only preoperatively but also intraoperatively.

Consent

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

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CRediT authorship contribution statement

Junji Iwasaki: The surgeon who performed the operation and follow-up, wrote the manuscript, performed the literature review, and provided approval of the final manuscript.
 Taku Iida: The surgeon who performed the operation and follow-up and provided approval of the final manuscript.
 Keiji Nagata: Supervision.
 Kojiro Nakamura: Supervision.

Table 1
Reported cases of duplicated cystic ducts draining a single gallbladder.

Case	Author/reference	Year	Age	Gender	Duplication type	Diagnosis
1	Perelman [3]	1961	56	Female	"H"	Intraoperative
2	Senapati et al. [4]	1984	56	Male	"Trabecular"	Intraoperative
3			55	Female	"H"	Intraoperative
4	Kubota [5]	1991	41	Female	"H"	ERCP
5	Nakasugi et al. [6]	1995	50	Female	"Y"	Preoperative ERCP
6	Ng et al. [7]	1996	60	Male	"H"	Intraoperative
7	Momiyama et al. [8]	1996	66	Female	"H"	Postoperative
8	Hirono et al. [9]	1997	74	Female	"H"	Intraoperative
9	Fujikawa et al. [1]	1998	70	Female	"H"	Intraoperative
10	Lobo et al. [10]	2000	49	Female	"Y"	Intraoperative
11	Tsutsumi et al. [11]	2000	74	Female	"H"	Preoperative ERCP
12	Shivhare et al. [12]	2002	46	Female	"H"	Intraoperative
13	Paraskevas et al. [13]	2007	76	Female	"H"	Preparation of cadavers
14	Huston et al. [14]	2008	43	Female	"H"	Intraoperative
15	Yoo et al. [15]	2008	55	Female	"H"	Intraoperative
16	Aristotle et al. [16]	2011	50	Male	"Y"	Preparation of cadavers
17	Shih et al. [17]	2011	37	Male	"Y"	Intraoperative
18	Shabanali et al. [18]	2014	50	Female	"H"	Intraoperative
19	Otaibi et al. [19]	2015	54	Male	"H"	Intraoperative
20	Samnani et al. [20]	2015	34	Female	"Y"	Intraoperative
21	Fujii et al. [21]	2017	57	Female	"Trabecular"	Preoperative MRCP, ERCP
22	Salih et al. [22]	2017	33	Female	"Y"	Intraoperative
23	Munie et al. [23]	2019	34	Female	"Trabecular"	Intraoperative
24	Bashian et al. [24]	2019	78	Male	"H"	Intraoperative
25	Harding et al. [25]	2020	78	Female	"H"	Intraoperative
26	Anisi et al. [26]	2020	58	Female	"Y"	Intraoperative
27	Abdelsalam et al. [27]	2021	42	Female	"Y"	Intraoperative
28	Lim et al. [28]	2021	82	Female	"Y"	Postoperative
29	Present case	2021	60	Female	"H"	Preoperative ERCP

Atsushi Itami: Supervision.

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Declaration of competing interest

The authors have no conflicts of interest to declare.

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