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A case of obstructive jaundice due to early carcinoma of the cystic duct protruding into the common bile duct

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ABSTRACT

INTRODUCTION: Cystic duct carcinoma is a rare disease, and only 33 cases reported worldwide have completely fulfilled the criteria first established by Farrar in 1951. Here we describe an extremely rare case of early cystic duct carcinoma that fulfilled the Farrar criteria, the papillary tumour protruding into the common bile duct, leading to obstructive jaundice.

CASE PRESENTATION: A 76-year-old man visited a clinic with icteric conjunctivae, and was referred to our hospital for investigation of suspected obstructive jaundice. He was initially diagnosed as having a distal bile duct carcinoma on the basis of ultrasonography (US), endoscopic retrograde cholangiopancreatography (ERCP), and magnetic resonance cholangiography (MRC), and underwent pancreatoduodenectomy with regional lymphadenectomy. Macroscopic examination showed that the stalk of the papillary tumour originated from the cystic duct, and that the protruding lesion was 50 mm in size. Histopathological examination revealed the tumour to be a papillary adenocarcinoma confined within the fibromuscular layer, with no evidence of lymph node metastasis. Therefore, the final diagnosis was early cystic duct carcinoma.

CONCLUSION: To our knowledge, this is the first case report of obstructive jaundice due to early carcinoma of the cystic duct protruding into the bile duct, with characteristics fulfilling the Farrar criteria.

1. Introduction

Primary carcinoma of the cystic duct is a rare disease. The cystic duct is a short and narrow tube that connects the gallbladder to the bile duct. In most cases, the origin of this malignancy is difficult to determine, which accounts for the rarity of reports. In 1951, Farrar proposed diagnostic criteria for cystic duct carcinoma. First, growth is restricted to within the cystic duct. Second, there must be no neoplastic process in the gallbladder or hepatic or common bile duct. Third, histological examination of growth must confirm the presence of carcinoma cells [1]. When we explored the previous cases by using data sources with "cystic duct carcinoma" and "Farrar criteria" as the search term beyond PubMed as well as Ichushi-Web from 1951 to 2017, only 33 cases were extracted. However, several cases of cystic duct carcinoma with invasion extending to the gallbladder neck or bile duct have been reported and classified as shown in Table 1. These new classifications considered tumour spread as well as invasion and would be more clinically useful [2–4] (Table1).

We have experienced a case of early carcinoma of the cystic duct, in which invasion was limited to the fibromuscular layer and the papillary tumour protruded into the common bile duct beyond the confluence of the cystic duct to reach the common bile duct, causing obstructive jaundice. Here we describe the clinical and pathological details of this case and discuss its rarity as well as the significant discrepancies from previous classifications that it exhibited. We also review the literature and summarize the presentation and management of this rare tumour. The work has been reported in line with the SCARE criteria [5].

Abbreviations: US, ultrasonography; ERCP, endoscopic retrograde cholangiopancreatography; MRC, magnetic resonance cholangiography; GOT, glutamate oxaloacetate transaminase; GPT, glutamate pyruvate transaminase; γ-GTP, γ-glutamyl transpeptidase; ALP, alkaline phosphatase; CEA, carcinoembryonic antigen; CA19-9, cancer antigen 19-9; ERBD, endoscopic retrograde biliary drainage; CT, computed tomography; PET, positron emission tomography; FDG, fluorodeoxyglucose; UICC, Union for International Cancer Control; GB, gallbladder; HE, hematoxylin and eosin; pap, papillary adenocarcinoma.

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Table 1
Comparison of the four different classifications.

Classification	Age (years)	Gender (n = M/F)	Total (n)	Jaundice (n)
Farrar (1951) 	60	21/12	33	5
Kim et al. (2007) Type Ⅰ Type Ⅱ Type Ⅲ 	ND	ND	20	ND
Yokoyama et al. (2008) Hepatic Hilum type Hepatic Hilum type Cystic Confluence type Cystic Confluence type 	ND	20/24	44	39
Nakata et al. (2009) Type I Type Ⅰ Type II Type Ⅱ Type III Type Ⅲ Type IV Type Ⅳ 	68	10/5	15	10

Abbreviations: ND: Data not mentioned in the report.
Farrar criteria: First, growth is restricted to within the cystic duct. Second, there must be no neoplastic process in the gallbladder or hepatic or common bile duct. Third, histological examination of growth must confirm the presence of carcinoma cells.
Classification by Kim et al.: Type I is confined to within the cystic duct. Type II means the tumor extends to the gallbladder neck or bile duct from the cystic duct side without obstructive jaundice. Type III indicates the tumor extends up to the gallbladder body or bile duct contralateral to the cystic duct opening, which then causes obstructive jaundice.
Classification by Yokoyama et al.: The hepatic hilum type means that the tumor mainly invades the hepatic hilum. The cystic confluence type indicates that the tumor invades the confluence of the cystic duct.
Classification by Nakata et al.: Type I means the tumor is located entirely within the cystic duct. Type II means that the tumor invasion has extended to the gallbladder. Type III means that the tumor invasion has extended to the common hepatic duct or common bile duct, including extension into the lumen and external invasion to the bile duct wall. Type IV means that the invasive lesion has extended to both the gallbladder and the bile duct.

2. Case presentation

A 76-year-old man visited a local clinic with icteric conjunctivae. He had sick sinus syndrome and used a pacemaker. Blood biochemistry revealed significantly high levels of total bilirubin and transaminase, and US imaging demonstrated intrahepatic bile duct dilatation. Therefore, he was referred to our department for examination of suspected obstructive jaundice.

On admission, the patient's body temperature was 35.9 °C, and yellowing of the conjunctivae and skin was evident. The patient had medium build, and no abnormal findings were evident in the neck or thoraco-abdominal region. Blood tests on admission showed no abnormality, but blood biochemistry revealed significant increases in the levels of transaminases and biliary enzymes (glutamate oxaloacetate transaminase (GOT): 260 U/L, glutamate pyruvate transaminase (GPT): 420 U/L, γ-glutamyl transpeptidase (γ-GTP): 1166 mU/mL, and alkaline phosphatase (ALP): 1163 U/L). The total bilirubin level was 6.0 mg/dL. Examination of tumour markers revealed a carcinoembryonic antigen (CEA) level of 3.0 ng/mL and a high level of cancer antigen 19-9 (CA19-9) (194.1 U/mL) (Table 2).

Endoscopic retrograde cholangiopancreatography (ERCP) revealed disruption of contrast medium flow from the confluence of the cystic and common hepatic ducts through the distal bile duct, as well as significant dilatation of the common and intrahepatic bile ducts. Therefore, an endoscopic retrograde biliary drainage (ERBD) stent was inserted for biliary drainage (Fig. 1a). Brush cytology at the site of distal bile duct stricture demonstrated class V (adenocarcinoma).

Table 2
Laboratory data.

Variable	Value
GOT (U/L)	260
GPT (U/L)	420
γ-GTP (mU/mL)	1166
ALP (U/L)	1163
T-Bil (mg/dL)	6.0
CEA (ng/mL)	3.0
CA19-9 (U/mL)	194.1

Abbreviations: GOT: glutamate oxaloacetate transaminase, GPT: glutamate pyruvate transaminase, γ-GTP: γ-glutamyl transpeptidase, ALP: alkaline phosphatase, T-Bil: total bilirubin, CEA: carcinoembryonic antigen, CA19-9: cancer antigen 19-9.

Abdominal computed tomography (CT) scan revealed a contrast-enhanced lesion that filled the lumen of the bile duct from inside the distal bile duct. This lesion did not extend beyond the walls of the bile duct, and neither infiltration into other organs nor no clear lymphadenopathy was observed (Fig. 1b). Positron emission tomography (PET)-CT scan revealed accumulation of fluorodeoxyglucose (FDG) that coincided with the lesion in the bile duct, and there were no clear findings of distant metastasis (Fig. 1c). Based on these results, we made a preoperative diagnosis of distal bile duct carcinoma (T1N0M0) according to the Union for International Cancer Control (UICC) classification and performed pancreatoduodenectomy with regional lymphadenectomy.