

[CASE REPORT]

Severe Acute Cholangitis and Bacteremia Due to *Campylobacter jejuni*: A Case Report and Review of the Literature

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Abstract:

Campylobacter jejuni is common cause of enteritis, but biliary infection rarely reported. An 82-year-old woman with pancreatic head cancer underwent endoscopic biliary drainage for malignant biliary obstruction. She was subsequently admitted for management of diarrhea. *C. jejuni* was identified in stool culture. Her symptoms resolved temporarily without antibiotics but flared up with a fever a few days later. She was diagnosed with acute cholangitis and bacteremia with *C. jejuni*. Endoscopic biliary drainage and antimicrobial administration improved her symptoms. As complications of *C. jejuni* diarrhea are rare, antibiotics are not necessarily indicated but sometimes are needed to prevent complications.

Key words: *Campylobacter jejuni*, acute cholangitis, *Campylobacter species*, *Campylobacter* enteritis

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Introduction

Campylobacter jejuni is a Gram-negative bacillus that may be curved or spiral-shaped and is a common cause of bacterial enteritis. Biliary infection caused by *Campylobacter* species rarely leads to bacteremia, but the risk of biliary infection is increased in patients who are elderly and those with underlying disease, such as cancer, human immunodeficiency virus infection, or diabetes.

We herein report a case of acute cholangitis and bacteremia following *C. jejuni* enteritis.

Case Report

An 82-year-old woman diagnosed with cancer of the head of the pancreas had undergone endoscopic biliary drainage and placement of a fully covered self-expandable metal stent with an antimigration system (Duckbill Biliary Stent,

Kawasumi Laboratory, Tokyo, Japan) (1) for malignant biliary obstruction 1 month earlier. She had declined antitumor treatment, including chemotherapy, and was being followed up in an outpatient clinic.

After the stent placement, she visited our hospital because of general weakness and a one-week history of persistent severe watery diarrhea. A stool sample was taken for culture, and probiotics were recommended. By the time the causative bacterial species was identified, the patient's symptoms had resolved. Therefore, no antibiotic treatment was provided. A few days later, she developed influenza A infection with a high-grade fever of approximately 39°C and generalized sharp jolts of pain due to physical contact. She was treated with oseltamivir phosphate, and the fever resolved temporarily but returned 15 days later, accompanied by abdominal pain and persistent watery diarrhea.

Laboratory tests showed elevated hepatobiliary enzyme levels (aspartate aminotransferase, 41 U/L; alkaline phosphatase, 574 U/L; gamma-glutamyl transpeptidase, 138 U/L).

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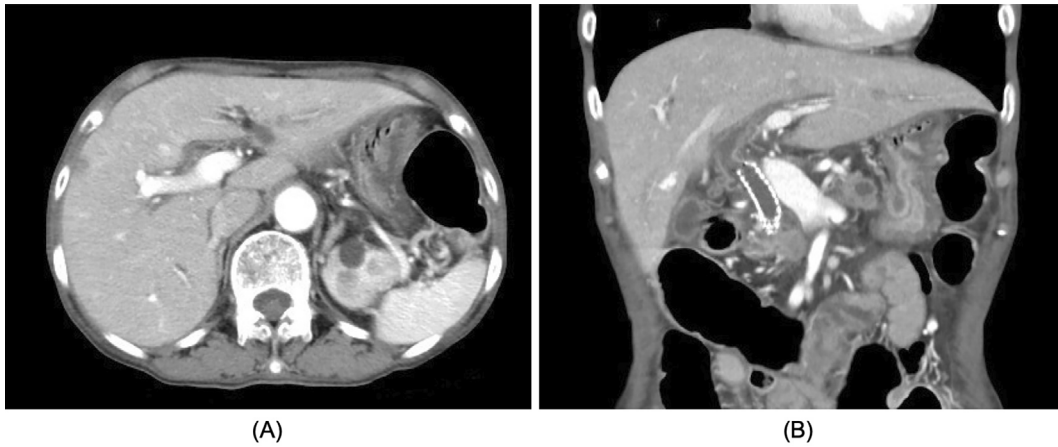


Figure 1. Abdominal contrast-enhanced computed tomography scans obtained at the onset of acute cholangitis. (A) Intrahepatic bile ducts are dilated. (B) The fully covered self-expandable metal stent is not obstructed and is appropriately positioned in the common bile duct. There was no wall thickening of the gallbladder or enlargement of the gallbladder.

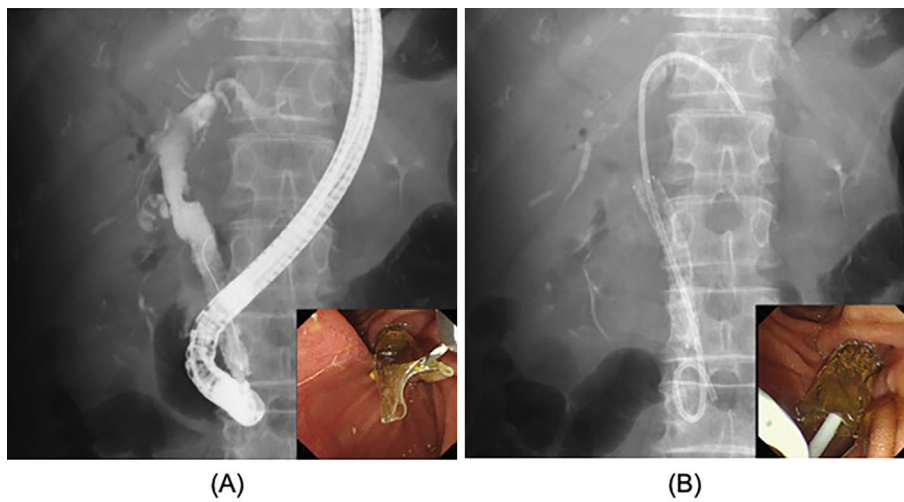


Figure 2. Endoscopic retrograde cholangiopancreatography for additional biliary drainage. (A) Cholangiography shows no stent dysfunction, such as obstruction or migration. (B) Endoscopy shows discharge of purulent bile from the self-expandable metal stent.

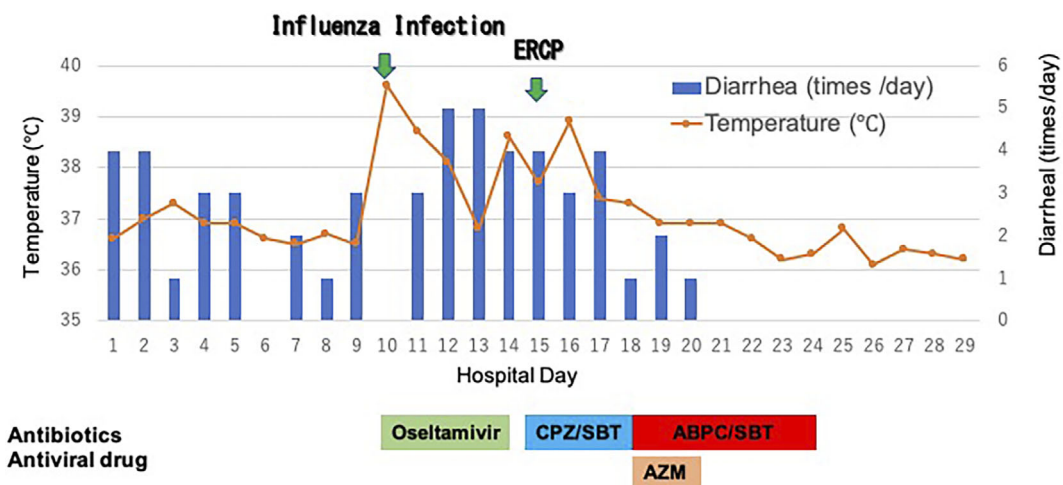


Figure 3. The patient's clinical course. ERCP: endoscopic retrograde cholangiopancreatography, CPZ/SBT: cefoperazone/sulbactam, ABPC/SBT: ampicillin-sulbactam, AZM: azithromycin and ampicillin-sulbactam

Table. Reports of Biliary Infection with *Campylobacter* Species.

Reference	Age (y)/ Sex	Biliary infection	Bacterial name	Preceding enteritis	Bacteremia	Underlying disease	Treatment
11	11/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	None	Antibiotics and operation
11	60/F	Acute cholecystitis	<i>C. jejuni</i>	No	None	Obstructive jaundice	Operation
11	32/F	Acute cholecystitis	<i>C. jejuni</i>	Yes	None	None	Operation
12	52/F	Acute cholecystitis	<i>C. jejuni</i>	Yes	None	Cholecystolithiasis, chronic cholecystitis	Antibiotics and operation
13	NA	Acute cholecystitis	<i>C. jejuni</i>	No	None	None	Operation
14	24/M	Acute cholecystitis	<i>Campylobacter</i> sp.	No	None	HIV	Antibiotics and operation
15	46/F	Acute cholecystitis	<i>Campylobacter</i> sp.	No	None	None	Operation
16	55/M	Acute cholecystitis	<i>Campylobacter</i> sp.	No	None	None	Antibiotics and operation
17	74/M	Acute cholangitis	<i>C. fetus</i>	No	None	Choledocholith	Antibiotics and endoscopic drainage
18	62/M	Acute cholecystitis	<i>Campylobacter</i> sp.	No	None	None	Antibiotics and operation
19	84/F	Acute cholecystitis	<i>C. jejuni</i>	Yes	None	None	Antibiotics and operation
20	59/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	None	Antibiotics and operation
21	83/M	Acute cholecystitis	<i>C. jejuni</i>	Yes	None	None	Antibiotics and operation
22	64/F	Acute cholecystitis	<i>C. fetus</i>	No	None	Hepatocellular carcinoma, obstructive jaundice	Antibiotics
23	62/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	Old myocardial infarction, hypertension	Antibiotics and operation
24	NA	Acute cholangitis	<i>C. fetus</i>	No	None	Immune deficiency syndrome	Antibiotics
25	51/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	Old myocardial infarction, hypothyreosis, dyslipidemia	Operation
26	35/M	Acute cholecystitis	<i>Campylobacter</i> sp.	Yes	None	None	Antibiotics and operation
27	71/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	Hypertension, old myocardial infarction	Antibiotics and operation
28	71/M	Acute cholangitis	<i>C. showae</i>	No	Existence	Biliary tract cancer	Antibiotics and endoscopic drainage
29	65/M	Acute cholecystitis	<i>C. jejuni</i>	No	None	Non-Hodgkin's lymphoma, cholelithiasis	Antibiotics
Our case	82/F	Acute cholangitis	<i>C. jejuni</i>	Yes	Existence	Cancer of the pancreatic head	Antibiotics and endoscopic drainage

C. jejuni: *Campylobacter jejuni*, *C. fetus*: *Campylobacter fetus*, *C. showae*: *Campylobacter showae*, HIV: human immunodeficiency virus

L), a low white blood cell count of 2,200/mm³, and a high C-reactive protein level of 3.1 mg/dL. Abdominal contrast-enhanced computed tomography showed dilated intrahepatic bile ducts with no wall thickening of the gallbladder or enlargement of the gallbladder (Fig. 1), as had been noted on previous computed tomography images. According to the 2018 Tokyo Guidelines, she was diagnosed with moderate acute cholangitis (2).

Endoscopic retrograde cholangiopancreatography showed that the stent was not obstructed but was discharging brownish, cloudy, purulent bile. The bile was cultured, a temporary plastic stent was placed for drainage (Fig. 2), and

the patient was started on cefoperazone-sulbactam for cholangitis. After the replacement of an additional stent, her abdominal symptoms improved rapidly. The blood culture subsequently identified *C. jejuni* and the bile culture identified *C. jejuni*, *Enterococcus casseliflavus*, and *Streptococcus anginosus*. The antimicrobial therapy was switched to intravenous azithromycin for 5 days and ampicillin-sulbactam for 7 days. Thereafter, the fever and diarrhea resolved (Fig. 3).

Discussion

This was a rare case of acute cholangitis where the causa-

tive organism was *C. jejuni*, which is a common cause of community-acquired bacterial enteritis. *C. jejuni* generally grows quite slowly, requiring 72-96 hours for primary isolation from stool samples and even longer from blood samples (3). Therefore, the symptoms and fever are likely to have already resolved by the time *C. jejuni* is identified in blood cultures from patients without underlying disease. As such, antibiotics are not always needed, but *C. jejuni* enteritis can become exacerbated, and bacteremia may develop as an extraintestinal complication, leading to acute biliary infection, pancreatitis, and peritonitis in rare cases (4).

Bacteremia occurs in 1% of *C. jejuni* infections (5-8), and biliary infection is very rare. Antibiotic therapy is needed in patients who are elderly, those who have serious underlying disease, and those who do not respond to conservative treatment (9, 10). In the present case, *C. jejuni* was identified on stool culture, and the infection appeared to resolve without antibiotics. A literature search revealed 23 cases of biliary infection caused by *Campylobacter* species in 21 reports (Table) (11-29). All cases caused by *C. jejuni* presented with acute cholecystitis, and all cases of acute cholangitis, except for our case, were caused by different species of the same genus. Most patients had an underlying biliary disorder, such as stones or malignancy, and a recent history of enteritis. Including our patient, there have been only two severe cases that led to bacteremia.

There are two possible pathways by which bacteremia could develop in patients with *C. jejuni* infection: the movement of *C. jejuni* from the intestinal tract into the biliary system and then into the blood vessels by cholangiovenous reflux (3) and direct bacterial translocation from the intestinal tract into the bloodstream independent of acute cholangitis (30). Given that the affinity of *C. jejuni* for vascular endothelium is lower than that of *C. fetus* (31), the *C. jejuni* bacteremia in our patient was likely induced by acute cholangitis. A self-expandable metallic stent with an antimigration system was used for biliary drainage under the expectation of long-term patency of the stent. Because of the rapid improvement in the patient's condition after placement of the additional biliary stent, the antimigration system may have interfered with biliary drainage and caused the severe bacteremia.

In conclusion, patients with an underlying biliary disorder and a history of *C. jejuni* enteritis should be monitored carefully for complications such as cholangitis and bacteremia.

The authors state that they have no Conflict of Interest (COI).

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