Case report

**Edwardsiella tarda-associated cholangitis associated with Lemmel syndrome**

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**ABSTRACT**

*Edwardsiella tarda* is an unusual human pathogen. Gastroenteritis is the most frequently reported manifestation of *E. tarda* infection and extraintestinal infection including cholangitis has rarely been reported. The overall mortality rate for *E. tarda* bacteremia is, however, reported to be up to 50% (Janda and Abbott, 1993). We describe a 80-year-old diabetic woman with cholangitis and *E. tarda* bacteremia with a biliary obstruction associated with a large juxtapapillary duodenal diverticulum (Lemmel syndrome) in the setting of past partial hepatectomy and cholecystectomy. She was successfully treated with endoscopic biliary drainage and antibiotics.

**Introduction**

*Edwardsiella tarda* is a rare human pathogen that can be associated with gastroenteritis. The extraintestinal manifestation include biliary tract infection, bacteremia, skin and soft tissue infection, liver abscess, peritonitis, intraabdominal abscess and tubo-ovarian abscess [2,3]. The major underlying diseases predisposing to *E. tarda* extraintestinal infection are hepatobiliary diseases, malignancy and diabetes mellitus [3].

This article describes a successful case of cholangitis accompanying bacteremia caused by *E. tarda* with diabetes mellitus, past history of operations of hepatobiliary systems and large juxtapapillary duodenal diverticulum.

**Case report**

An 80-year-old woman was admitted to our hospital with fever of 5 days duration. She had a history of hepatocellular carcinoma and gallbladder cancer, which had been successfully treated by partial hepatectomy and cholecystectomy, as well as diabetes mellitus and mild interstitial pulmonary fibrosis. She did not smoke or consume any alcohol and was taking daily oral aspirin for a cerebrovascular accident prevention.

The physical examination showed temperature of 38 °C, heart rate 102 and blood pressure 111/67 and she had icteric sclerae. Her breath sounds and heart sounds were normal and she had no rash or lymphadenopathy. Her abdomen was soft and nontender. The laboratory data showed a white blood cell count of 9210/μL, platelet count 185,000/μL, hemoglobin 15.7 g/dL, CRP 19.7 mg/dL and blood glucose 347 mg/dL. Her LDH was 348 IU/L, AST 259 IU/L, ALT 233 IU/L, total bilirubin 6.2 mg/dL, GGTP 1734 IU/L, alkaline phosphatase 1200 IU/L, amylase 563 IU/L (74% pancreatic), lipase 843 IU/L and she had pyuria (30–49 WBC/high power field).

Contrast-enhanced computed tomography (CT) showed dilation of common and intrahepatic bile duct and main pancreatic duct, juxtapapillary duodenal diverticulum, heterogeneous early stain in liver parenchyma on arterial phase. No apparent bile duct stones were found (Fig. 1).

She was diagnosed as having cholangitis of moderate grade and got emergently endoscopic retrograde cholangiopancreatography. During that procedure, it was unable to visually recognize major duodenal papilla because of large juxtapapillary duodenal diverticulum with food residue that was difficult to remove. The proximal side of major duodenal papilla was cut and cannulated to the exposed common bile duct (Fig. 2a,b). It was felt that the cholangitis occurred due to biliary obstruction from a juxtapapillary duodenal diverticulum, so-called Lemmel syndrome and an indwelling endoscopically biliary drainage tube was placed (Fig. 2c) and cefazolin was administered as antimicrobial therapy. The culture of venous blood and bile duct fluid both revealed *E. tarda* which was sensitive to almost all antimicrobials (Table 1). She improved and was discharged on the 13th hospital day.

**Discussion**

*Edwardsiella tarda*, a member of the family *Enterobacteriaceae*, is a rare human pathogen. This organism has been isolated predominantly in freshwater, marine environment, and in animals living in such
environments [3]. So contact with these animals and consumption of contaminated food, e.g. sushi, raw fish is considered as risk factors of *E. tarda* infection [4–6]. It seldom colonizes in the human gastrointestinal tract, reported to be found only in 26 of 353,600 Japanese individuals [7,8]. Healthy carrier rate of *E. tarda* is only 0.007% in Japan and it is detected generally during May and November, especially July and August [7]. The extraintestinal manifestations include biliary tract infection, bacteremia, skin and soft tissue infection, liver abscess, peritonitis, intraabdominal abscess and tubo-ovarian abscess [3]. Sepsis caused by *E. tarda* is said to develop in immunocompromised hosts and patients with biliary disease, malignancy and diabetes mellitus, and reported mortality of approximately 50% [1].

This episode occurred in November and the source of the *E. tarda* was not identified although the consumption of contaminated food was...
suspected as the origin. The route was thought to be retrograde from the duodenum rather than hematogenous route via the portal vein and that the duodenal diverticulum served as the main inciting cause of the biliary obstruction and subsequent cholangitis. In patients with a large juxtapapillary duodenal diverticulum papillitis can be induced by chronic diverticular inflammation [9] causing papillary fibrosis and stenosis, by direct obstruction of common bile duct by the diverticulum or its impacted material [10] and by dysfunction of the papilla are considered as mechanisms of obstruction and subsequent cholangitis [11].

Generally, *Edwardsiella* tarda is susceptible to a wide range of antibiotics. Also in our case, *E.tarda* was sensitive to almost all antibiotics except amikacin.

### Table 1

| Antibiotics | MIC  | Sensitivity |
|-------------|------|-------------|
| cefazolin   | 0.5  | sensitive   |
| ceftriaxone | ≤ 0.06 | sensitive |
| ceftazidime | ≤ 0.06 | sensitive |
| cefepime    | ≤ 0.06 | sensitive |
| cefepimepiperacillin | ≤ 0.25 | sensitive |
| imipenem/cilastatin | 0.5 | sensitive |
| meropenem   | ≤ 0.03 | sensitive |
| levofloxacin| ≤ 0.03 | sensitive |
| cefuroxime  | ≤ 0.03 | sensitive |
| amikacin    | 2    | resistant   |
| tobramycin  | 1    | sensitive   |
| minocycline | ≤ 0.06 | sensitive   |

**Conclusion**

We report a rare case of *E. tarda* cholangitis accompanying bacteremia with juxtapapillary duodenal diverticulum treated successfully with antimicrobials and endoscopic biliary drainage.

**References**

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