CASE REPORT

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Spontaneous Choledochoduodenal Fistula after Metallic Biliary Stent Placement in a Patient with Ampulla of Vater Carcinoma

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Biliary stent-related enteric perforations are very rare complications that are caused by the sharp end of a metallic stent, stent migration, or tumor invasion. Moreover, the choledochoduodenal fistula resulting from metallic biliary stent-induced perforation is extremely rare. Here, we report a case in which a spontaneous choledochoduodenal fistula occurred after biliary metallic stent placement in a patient with an Ampulla of Vater carcinoma but was successfully managed by supportive treatments, including nasobiliary drainage. This case might have occurred as the result of a rupture of the bile duct following pressure necrosis and inflammation caused by impacted calculi and food materials over the tumor ingrowth in the uncovered biliary stent. **(Gut and Liver 2009;3:360-363)**

Key Words: Choledochoduodenal fistula; Stents; Ampulla of Vater

INTRODUCTION

As biliary stents are used frequently and for long periods of time, stent-related complications are increasing.[1,2] Among biliary stent-related intestinal complications, the choledochoduodenal fistula resulting from metallic biliary stent-induced perforation is a very rare complication.

To our knowledge, this is the first report of a spontaneous choledochoduodenal fistula as a delayed complication after biliary metallic stent placement in a patient with an Ampulla of Vater (AOV) carcinoma. The complication was successfully managed by supportive treatments that included nasobiliary drainage.

CASE REPORT

A 76-year-old woman presented with a severe pain in the right upper quadrant of the abdomen and with jaundice that had started to worsen four days before. An uncovered biliary stent (Niti-S, biliary uncovered stent; Taewoong, Seoul, Korea) that was 5 cm in length had been inserted 40 days earlier for palliative management of an AOV carcinoma due to refusal of a curative surgical or endoscopic resection, despite the T1-stage tumor. Laboratory tests revealed the following: white blood cell count, 14.380×10^9/L ($4.0\text{-}10.8\times10^9$/L); total bilirubin, 15.2 mg/dL (0.2-1.2 mg/dL); AST, 75 IU/L (0-40 IU/L); ALT, 72 IU/L (0-40 IU/L); and alkaline phosphatase, 225 IU/L (39-117 IU/L).

Abdomen computed tomography (CT) showed an approximately 2 cm diameter cystic lesion between the duodenum and the mid-portion of the metallic stent, as well as upstream bile duct dilation (Fig. 1). The following day, the patient's abdominal pain spontaneously improved, and total bilirubin decreased from 15.2 mg/dL to 10.8 mg/dL. A subsequent endoscopy demonstrated that the bile had drained through a small fistula on the medial wall of the proximal second portion of the duodenum (Fig. 2A). The endoscopy also showed that the stent was impacted with food materials and stones (Fig. 2B). However, there was no stent migration or deformation on the end of the stent. When a mixture of indigo carmine and contrast

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was injected into the common bile duct, the contrast mixture was passed out the duodenum via the same fistula opening (Fig. 2C). As the dye was injected through an endoscopic nasobiliary drainage (ENBD) tube, fluoroscopy showed a linear track of the fistula from the mid-portion of the metallic stent to the duodenum, which was dissimilar to the CT finding. There was no evidence of contrast leakage into the extraduodenal abdominal cavity (Fig. 3A).

Seven days later, after placement of an ENBD tube and conservative management, including antibiotics, there was no fistula found from follow-up cholangiography (Fig. 3B). There was no evidence of tumor invasion on the fistula opening upon pathological examination.

Following the removal of several impacted stones, a covered biliary stent (Niti-S, biliary covered stent; Taewoong) that was 6 cm in length was reinserted for palliative management of the tumor ingrowth and fistula. The patient remained under outpatient observation without recurrence for the following 3 months.

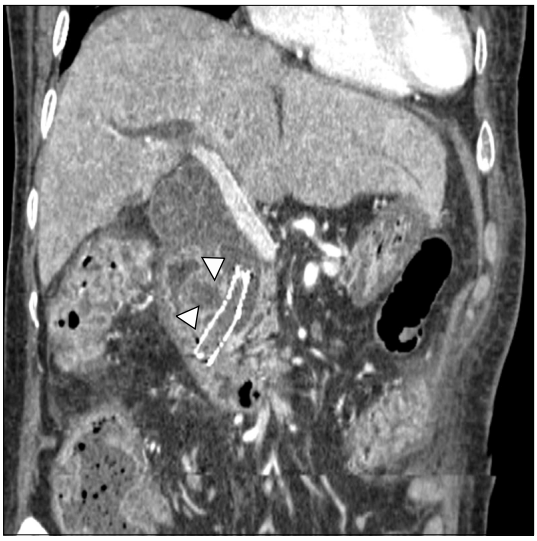

Fig. 1. Abdomen CT shows a round cystic lesion between the duodenum and the mid-portion of a metallic stent in a common bile duct (arrow head).

DISCUSSION

Biliary stent-related enteric perforations are very rare complications, with an incidence below 1%. Intestinal perforations are usually caused by the sharp end of a metallic stent or stent migration.[1-3] Choledochoduodenal fistula caused by tumor invasion or spontaneous perforation of the bile duct have rarely been reported.[4-6]

Spontaneous biliary-enteric fistulas have an incidence rate of 3-5% among cases; they are usually caused by calculi in the inflamed gallbladder or biliary tract, peptic ulcers, trauma, Crohn's disease, or malignant invasion by an AOV carcinoma or cholangiocarcinoma.[4-11] Among these causes, a choledochoduodenal fistula associated with an AOV carcinoma is an extremely rare complication, and metallic biliary stent-related spontaneous choledochoduodenal fistulas without tumor invasion have not yet been reported.

This unusual complication developed spontaneously in a patient with an AOV carcinoma as a delayed complication after metallic biliary stent placement. However, there was no evidence of tumor invasion at the fistula

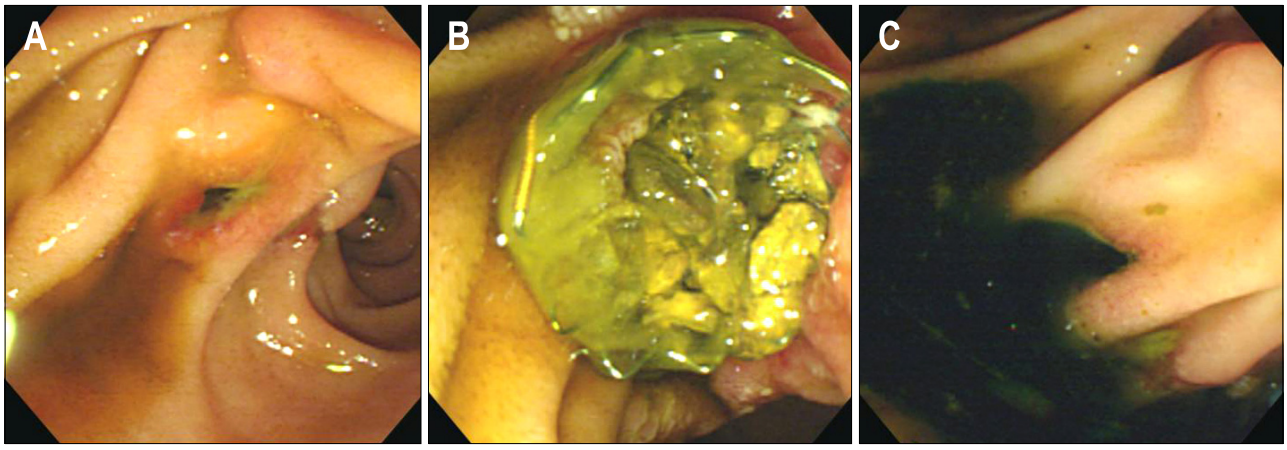

Fig. 2. (A) Endoscopic findings demonstrate that the bile drained through a small fistula on the medial wall of the proximal second portion of the duodenum. (B) The occluded metallic biliary stent. (C) A mixture of indigo carmine and contrast flow out via the fistula opening.

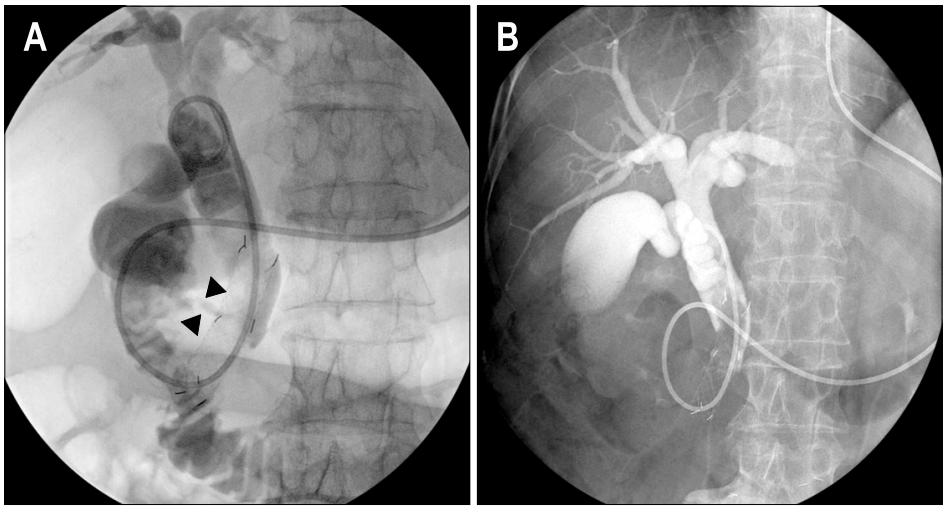

Fig. 3. (A) Cholangiography reveals the linear track of the fistula from the mid-portion of the metallic stent to the duodenum, without contrast leakage (arrow head). (B) Follow-up cholangiography does not show the fistula tract.

site, and no evidence of metal stent migration or deformation of the stent end at endoscopic or radiologic findings. The cystic formation between the CBD and duodenum appeared to have been precipitated by a rise in intraductal pressure, following obstruction by distal bile duct stones and tumor ingrowth in the uncovered biliary stent. Subsequently, cystic rupture in the duodenum created a choledochoduodenal fistula. The spontaneous symptomatic and laboratory improvement, along with the linear track formation of the fistula from the mid-portion of the metallic stent to the duodenum, which was dissimilar to the CT finding of a round cystic lesion, were suspected to have resulted from the rupture of an inflammatory cystic lesion in the duodenum. By excluding the possibilities of tumor metastasis, direct stent migration and deformation, we differentiated those causes and suggested the pathological explanation that rising intraductal pressure induced necrosis of the duct wall due to direct erosion and inflammation by stones or an uncovered stent.

Management of choledochoduodenal fistulas depends on their type and etiology. In fistulas with complicating duodenal ulcers, medical management or surgery can be used. Endoscopic management and the extraction of a bile duct stone (if present), may be needed.[5,7,8,12,13] There is a justified trend towards conservative management of iatrogenic bile duct perforations.[2] The current case was also managed conservatively, by reducing the pressure gradient with nasobiliary drainage and antibiotics.

In summary, we have been successful in conservatively managing a spontaneous choledochoduodenal fistula that was precipitated by a rise in intraductal pressure following obstruction by distal bile duct stones and tumor ingrowth in the uncovered biliary stent.

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