Ampullary Cancer with Pancreas Divisum Treated by Endoscopic Partial Papillectomy: a Case Report

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Abstract

We herein report a case of ampullary cancer associated with pancreas divisum. Duodenoscopy revealed a tumor at the ampulla of Vater with a normal ampullary orifice. Intraductal ultrasonography showed a hypoechoic mass limited to the ampulla of Vater. ERCP showed an arborizing ventral pancreatic duct without connection to the dorsal duct. Endoscopic snare resection of the tumor was performed following biliary stenting. Histological examination revealed well-differentiated tubular adenocarcinoma limited to the ampulla of Vater. Endoscopic papillectomy with the assistance of a biliary stent is useful in cases of ampullary neoplasm with a normal ampullary orifice in order to avoid ductal injury.

Key words

Ampullary neoplasm – ampulla of Vater – ERCP – biliary stenting – pancreatic duct stent – endoscopic papillectomy

Introduction

Endoscopic papillectomy has been reported to be useful for adenomas of the ampulla of Vater [1, 2]. This technique has been suggested as an alternative to surgery in the treatment of cancer of the ampulla of Vater limited to the mucosa or its components [3, 4]. We herein report a rare case of ampullary cancer associated with pancreas divisum which was successfully treated by endoscopic partial papillectomy.

Case report

A 62-year-old man who had undergone surgical treatment for rectal cancer two months previously underwent esophago-gastro-duodenoscopy (EGD) for a general check-up, which led to detection of a tumor at the ampulla of Vater. He was referred to our department for further evaluation. He had no history of familial adenomatous polyposis. Laboratory data, including serum CEA and CA19-9, revealed no abnormalities. Ultrasound examination findings were normal. Duodenoscopy revealed a reddish exposed-type tumor mass at the ampulla of Vater with a normal ampullary orifice (Fig. 1). Endoscopic ultrasonography (EUS) demonstrated a hypoechoic mass, measuring 20 mm in size, at the ampulla of Vater. The tumor echo was limited to the ampulla of Vater, without invasion into the duodenal muscularis propria layer or pancreatic parenchyma. CT revealed no distant metastasis nor dilation of the bile duct. MR cholangiopancreatography could not be performed due to the presence of a metal in his body. Endoscopic retrograde cholangiopancreatography (ERCP) showed an arborizing ventral pancreatic duct without connection to the dorsal duct (Fig. 2). Transpapillary intraductal ultrasonography (IDUS) of the bile duct showed a hypoechoic mass limited to the...
ampulla of Vater without invasion of the duodenal muscularis propria layer or pancreatic parenchyma. Well-differentiated tubular adenocarcinoma was highly suspected by the biopsy of the tumor.

Based on the diagnosis of T1 stage ampullary adenocarcinoma according to the TNM classification [5] an endoscopic papillectomy was performed after informed consent was obtained from the patient. At first, using a duodenoscope (JF-260V: Olympus Medical Systems, Co., Ltd. Tokyo, Japan), a stent (Flexima®: Microvasive, Boston Scientific Corp., Natick, MA) was placed in the bile duct in order to avoid biliary injury (Fig. 3a). Without local injection into the submucosal layer, tumor excision was successfully performed by using a snare 20 mm in size with the 120-watt Endocut® mode (ICC 200: ERBE Corp., Tuebingen, Germany) for about 2 seconds above the stent, without evidence of the residual tumor (Fig. 3b). Transpapillary pancreatic duct stenting to prevent post-ERCP pancreatitis was not carried out. There were no clinical symptoms suggesting development of cholangitis/pancreatitis. Oral feeding was commenced two days after the procedure without any adverse events. The biliary stent was removed eight days after the procedure.

Histology of the resected specimen, 18 mm in size, revealed well-differentiated tubular adenocarcinoma with low-grade cytological atypia (Fig. 4), which was limited to the mucosa, with lymphoid stroma. The tumor cells showed high Ki-67 labeling index (about 30%) and were conspicuously positive for chromogranin A and serotonin. The margin of the resected specimen was free of tumor at both the horizontal and vertical ends. Neither lymphatic permeation nor vascular invasion were demonstrated. The final histological stage of the tumor was T1.

**Discussion**

Endoscopic papillectomy for ampullary adenoma has been reported since the early 1980s [1, 2]. For ampullary cancer, pancreaticoduodenectomy has been considered to be the standard treatment. However, some reports have
suggested endoscopic treatment as an alternative to surgery for selected patients with ampullary cancer [3, 4]. Therefore, accurate preoperative staging is mandatory for making therapeutic decisions. EUS and IDUS play an important role in this context [6]. Based on the pathological findings of the resected specimen [3] the indications for endoscopic papillectomy in patients with ampullary cancer at our institution are T1 cancer limited to the mucosal layer and no spread to the bile duct and the pancreatic duct.

Pancreatic duct stenting after endoscopic papillectomy can lessen the frequency of post-ERCP pancreatitis in patients with ampullary neoplasm [7]. However, the diagnosis of pancreas divisum was made based on the ERCP findings in this particular case. Since the minor papilla of Vater drained pancreatic juice from the dorsal pancreas, transpapillary pancreatic duct stenting was not essential to prevent post-ERCP pancreatitis.

Since the present case had a normal ampullary orifice without tumor invasion, it was not necessary to resect the whole papilla of Vater including the ampullary orifice. Tumor excision was successfully performed above the stent placed in the bile duct without any adverse events, resulting in a histologically cancer-free margin of the resected specimen. The placement of a biliary stent was suggested as contributing to the prevention of a biliary injury.

Based on the PubMed database and Japanese literature database ‘Igaku-Chuou Zashi’ with the key words ‘ampullary cancer’ and ‘pancreas divisum’, only two cases of ampullary cancer with pancreas divisum have been reported since 1983 up to 2009 [8, 9]. Okamoto et al [8] reported a case of ampullary carcinoma, 11 mm in size, with pancreas divisum who underwent pancreaticoduodenectomy. The resected specimen of their case was histopathologically diagnosed as T1. Kobayashi et al [9] also reported a case of ampullary cancer (30 mm in size, T3 stage) with pancreas divisum where a pancreaticoduodenectomy was performed. The present case is the first case of ampullary carcinoma, 11 mm in size, with pancreas divisum who underwent pancreaticoduodenectomy. The placement of a biliary stent was suggested as contributing to the prevention of a biliary injury.

In conclusion, the present case is the first case of ampullary cancer with pancreas divisum at the ampulla of Vater, treated by endoscopic partial papillectomy. A biliary stent placement was used to avoid ductal injury at endoscopic resection.

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References