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Adenocarcinoma of the Minor Duodenal Papilla: Report of a Case

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Key Words
Minor duodenal papilla · Adenocarcinoma · Accessory pancreatic duct

Abstract
An 81-year-old male was found to have a duodenal tumor by screening upper gastrointestinal endoscopy. The tumor was located in the minor duodenal papilla. Pathological examination of the biopsy specimen revealed adenocarcinoma, and endoscopic ultrasound showed an elevated hypoechoic mass in the minor duodenal papilla. The preoperative diagnosis was therefore considered to be either adenocarcinoma of the minor duodenal papilla or duodenal cancer. We performed a subtotal stomach-preserving pancreaticoduodenectomy. Histopathological examination of the resected specimen showed the tumor cells to be primarily located in the submucosa of the minor duodenal papilla, with slight invasion into the pancreatic parenchyma through the accessory pancreatic duct. We therefore diagnosed a primary adenocarcima of the minor duodenal papilla. Adenocarcinoma of the minor duodenal papilla is considered to be a rare disease, but it may be underestimated because of the difficulty in distinguishing advanced adenocarcinoma of the minor duodenal papilla from primary duodenal cancer and cancer of the pancreatic head.

Introduction

Either the minor duodenal papilla, which is the orifice of the accessory, or the dorsal pancreatic duct (Santorini duct), which is mostly accompanied by pancreatic tissue, is situated in the 2nd portion of the duodenum, typically about 2 cm ventroproximal to the major duodenal papilla [1]. Tumors in the minor duodenal papilla are considered to be rare neoplasms [2]. In particular, adenocarcinoma of the minor duodenal papilla is

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considered to be an extremely rare disease, and only five cases of this disease have been previously reported [2–6]. The reasons for this scarcity are attributed to (1) the low incidence of this disease, (2) the fact that there are no specific symptoms, and (3) the difficulty in distinguishing advanced adenocarcinoma of the minor duodenal papilla from primary duodenal cancer and cancer of the pancreatic head [4]. We herein report a patient with adenocarcinoma of the minor duodenal papilla who was diagnosed by screening upper gastrointestinal endoscopy and was thereafter treated successfully with surgical treatment. Moreover, we also review the pertinent literature and discuss the clinical characteristics, pathological investigation and treatment options.

Case Report

An 81-year-old male was admitted to our hospital because his general practitioner had performed routine screening upper gastrointestinal endoscopy and found an irregular elevated tumor in the 2nd portion of the duodenum. He had a past history of transurethral resection of the prostate due to prostate hypertrophy. We performed upper gastrointestinal endoscopy and found the tumor to be located 2 cm proximal to the major duodenal papilla where the minor duodenal papilla should have been (**fig. 1**). Pathological examination of a biopsy specimen of this tumor revealed the presence of papillary adenocarcinoma. Laboratory examinations revealed an elevated CA19-9 level (100.1 U/ml: normal range <37), but all other findings, including hematological profile, renal function, pancreatic enzymes, liver enzymes, electrolytes, and CEA were within the normal range.

Computed tomography (CT) was not able to demonstrate a primary tumor of the duodenum and revealed no apparent distant metastasis, lymph node metastasis, or peritoneal dissemination. Endoscopic ultrasound (EUS) showed an elevated hypoechoic mass in the minor duodenal papilla with retention of the muscularis propria of the duodenum (**fig. 2**). Based on these findings, the most probable preoperative diagnosis was carcinoma of the minor duodenal papilla or duodenal cancer.

We performed a subtotal stomach-preserving pancreaticoduodenectomy. At laparotomy, there was no liver metastasis or peritoneal dissemination. A hard mass, which might have invaded the pancreas, was palpable in the 2nd portion of the duodenum. After subtotal stomach-preserving pancreaticoduodenectomy, reconstruction by the modified Child method was done in the following order: pancreaticojejunostomy, hepaticojejunostomy and gastrojejunostomy. Histopathological examination of the resected specimen identified the tumor to consist of papillary adenocarcinoma, well differentiated tubular adenocarcinoma and moderately differentiated tubular adenocarcinoma (**fig. 3**a). The tumor cells were primarily located in the submucosa of the minor duodenal papilla, which consists of the pancreatic tissue of the dorsal pancreas, accessory pancreatic duct and the surrounding fibrous connective tissue, with a slight degree of invasion into the pancreatic parenchyma through the accessory pancreatic duct (fig. 3b, c). There was no finding of lymph node metastasis. Based on these pathological findings, we eventually diagnosed primary adenocarcima of the minor duodenal papilla. In addition, histopathological examination accidentally revealed a 4 mm tumor, which was diagnosed to be a gastrointestinal stromal tumor of the duodenal bulb.

The patient's postoperative course was uneventful, and he was discharged 17 days after surgery without the need for insulin injections. Postoperative adjuvant therapy was not scheduled because of the early stage of the disease.

Discussion

The minor duodenal papilla, which is occasionally difficult to distinguish macroscopically, can be identified in virtually all cases. The minor duodenal papilla is located in the anterior wall of the 2nd portion of the duodenum, about 2 cm proximal to the major duodenal papilla [7], and consists of (1) the accessory pancreatic duct passing through the muscularis propria of the duodenum, (2) the pancreatic tissue of the dorsal

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pancreas, which is continuous with the proper pancreas parenchyma in about 40% of cases, and (3) the surrounding fibrous connective tissue [1]. Neoplasms of the minor duodenal papilla are rare, and most reported cases have been found to be benign tumors, such as carcinoid tumors and adenomas. Adenocarcinoma of the minor duodenal papilla is considered to be an extremely rare disease, and only five cases of this disease have been reported previously (**table 1**) [2–6]. However, the incidence of this disease may be underestimated because of the difficulty in distinguishing advanced adenocarcinoma of the minor duodenal papilla from primary duodenal cancer and cancer of the pancreatic head [1]. In other words, most of the previously reported cases of adenocarcinoma of the minor duodenal papilla were discovered incidentally at a relatively early stage by screening upper gastrointestinal endoscopy, and as a result, the patients developed no characteristic symptoms [4]. According to the reported cases in the pertinent literature, for the evaluation of tumor staging, CT and EUS for tumors of the minor duodenal papilla are considered to be useful [5]. A diagnosis of whether or not the tumor has infiltrated into the pancreatic parenchyma is an important matter and a predictive factor of prognosis.

Our patient also had no symptoms, and the tumor located 2 cm proximal to the major duodenal papilla was identified by screening upper gastrointestinal endoscopy. To determine the invasion depth of the tumor, CT and EUS were performed. CT was not able to demonstrate a primary tumor of the minor duodenal papilla, but EUS revealed a hypoechoic mass with slight retention of the muscularis propria of the duodenum. These EUS findings were consistent with the pathological findings of the resected specimen.

Primary adenocarcinoma of the minor duodenal papilla is histopathologically defined as a tumor derived from the epithelium covering the minor papilla, the pancreatic tissue of the dorsal pancreas and the accessory pancreatic duct within the minor papilla [4]. All five previously reported cases had a histopathological classification of either well differentiated adenocarcinoma or moderately differentiated adenocarcinoma, and none of the cases had lymph node metastasis. In our case, which was diagnosed to be papillary adenocarcinoma, well differentiated tubular adenocarcinoma and moderately differentiated tubular adenocarcinoma were observed to proliferate in the submucosa of the minor duodenal papilla, and there was also slight invasion into the pancreatic parenchyma through the accessory pancreatic duct, but no lymph node metastasis was observed. These findings (differentiation status, lack of lymph node involvement) in the reported cases may therefore be characteristic of primary adenocarcinoma of the minor duodenal papilla, or may merely be associated with the early detection of the disease. Although it remains unclear whether this tumor originated from the epithelium covering the minor duodenal papilla or the accessory pancreatic duct, it was classified as a primary adenocarcinoma of the minor duodenal papilla.

For the treatment of primary adenocarcinoma of the minor duodenal papilla, pancreaticoduodenectomy has so far been performed for all reported cases. Considering that tumors of the minor duodenal papilla may spread to the duodenum or the pancreatic parenchyma and thereby possibly result in lymph node metastasis, pancreaticoduodenectomy is thought to be a justified procedure. In our case, EUS showed a tumor with slight retention of the muscularis propria of the duodenum, and there was a possibility that the tumor might spread to the pancreatic parenchyma, therefore we performed pancreaticoduodenectomy. Recently, endoscopic resection (papillectomy) for either adenoma or carcinoid tumors of the minor duodenal papilla has been reported, and

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few complications and either no or only minimal residual tumors have been observed [8–10]. However, a patient who demonstrates tumor invasion into the surrounding tissue or lymph node metastasis is considered to be contraindicated for endoscopic resection.

Conclusion

We herein describe a patient presenting with adenocarcinoma of the minor duodenal papilla, which is considered to be an extremely rare disease. No consensus exists yet in regard to both the primary treatment as well as the optimal adjuvant chemotherapy and the prognosis of adenocarcinoma of the minor duodenal papilla, and therefore further accumulation of similar cases and continued investigation of this disease are necessary.

Table 1. Reported cases of adenocarcinoma of the minor duodenal papilla

Year	First author	Age	Sex	Chief complaint	Treat-ment	Size (mm)	Pathology	Tumor localization	Lymph node metastasis	Major papilla	Divism
1998	Yamao	77	male	transient epigastric pain	PpPD	25×20	mod.	duodenal mucosa to pancreatic parenchyma	no	normal	no
2007	Kajiwara	60	male	transient abdominal pain	SSpPD	50×30	well	duodenal mucosa	no	normal	yes
2008	Wakatsuki	70	male	no	PpPD	11×8	well	duodenal submucosa	no	normal	no
2008	Parthasarathy	60	female	fever and jaundice	PD	15×12	mod.	major papilla: invasion into pancreas and duodenum; minor papilla: unkown	no	adenocarcinoma	un-known
2008	Matheus	50	female	abdominal pain and jaundice	PpPD	10	mod.	major papilla: invasion into duodenum; minor papilla: unkown	no	adenocarcinoma	un-known
2011	Our case	81	male	no	SSpPD	20×15	pap. + well + mod.	duodenal submucosa with slight invasion into the pancreatic parenchyma	no	normal	no

PpPD = Pylorus-preserving pancreaticoduodenectomy; SSpPD = subtotal stomach-preserving pancreaticoduodenectomy; PD = pancreaticoduodenectomy.

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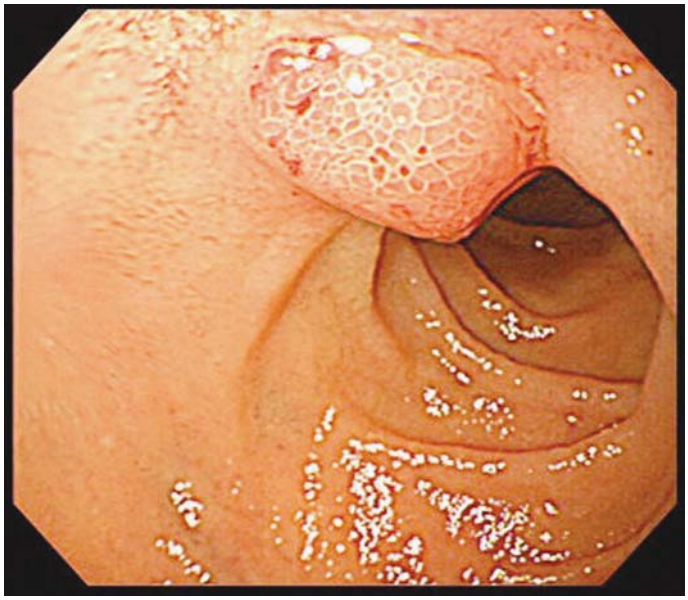

Fig. 1. Upper gastrointestinal endoscopy showed an irregular elevated tumor, which was located 2 cm proximal to the major duodenal papilla (where the minor duodenal papilla should have been), thereby revealing a normal major duodenal papilla. Biopsy results of this tumor indicated papillary adenocarcinoma.

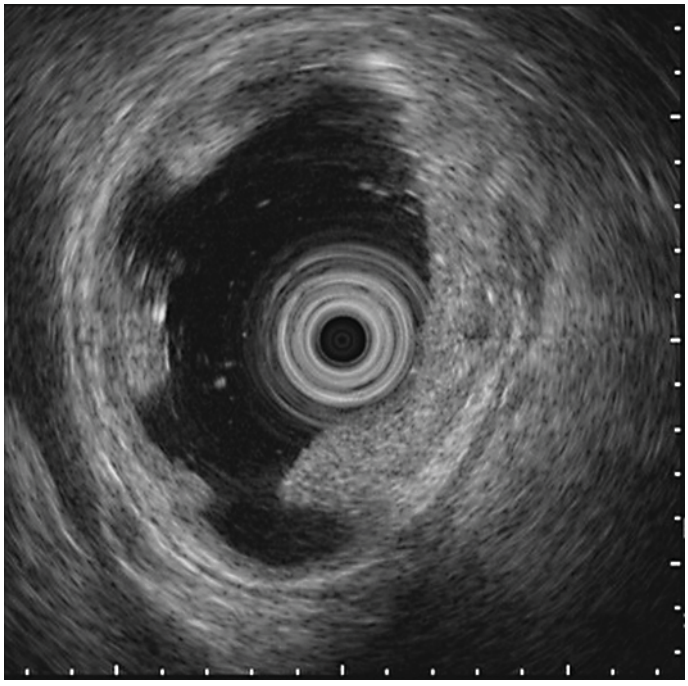

Fig. 2. EUS revealed an elevated hypoechoic mass in the minor duodenal papilla. According to the EUS findings, the layer of the muscularis propria was interrupted. As a result there was a possibility that the tumor might spread to both the muscularis propia of the duodenum and pancreatic parenchyma.

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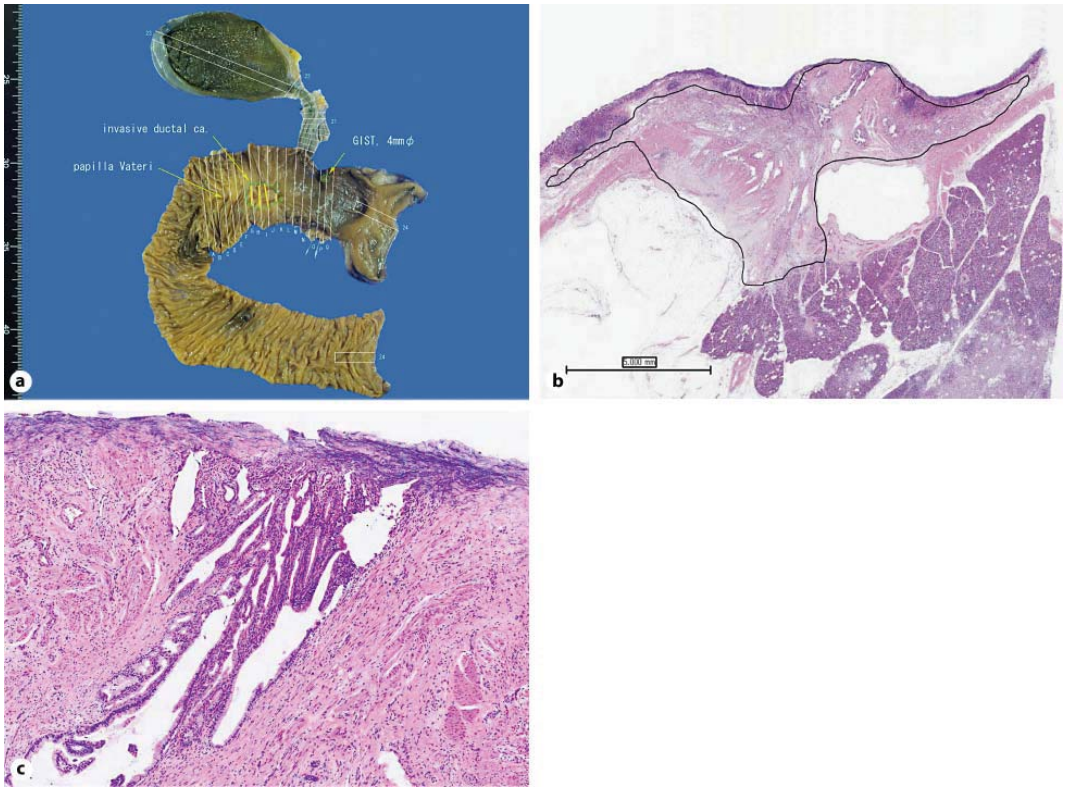

Fig. 3. **a** A resected specimen of the duodenum showed an irregularly elevated tumor (adenocarcinoma) measuring 20 × 15 mm in the minor duodenal papilla, and a submucosal tumor (gastrointestinal stromal tumor) measuring 4 mm in the duodenal bulb. The major duodenal papilla was normal in both size and shape. **b**, **c** Tumor cells were primarily located in the submucosa of the minor duodenal papilla, and there was slight invasion into the pancreatic parenchyma through the accessory pancreatic duct. These findings indicate that this tumor originated from either the minor duodenal papilla or an accessory pancreatic duct.

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