

Adenocarcinoma of the Minor Duodenal Papilla: Report of a Case

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Abstract

We report a rare case of adenocarcinoma of the minor papilla of the duodenum treated with transduodenal minor papillectomy. A 64-year-old woman was treated for an asymptomatic duodenal tumor detected on gastroduodenoscopy. Endoscopy showed a 15-mm sessile mass in the descending duodenum proximal to the major papilla. The major papilla was a villous 24-mm-diameter polypoid tumor. Histopathologic examination of the biopsy specimen showed tubular adenoma with moderate epithelial atypia. Transduodenal major and minor papillectomies were performed. The orifice of the duct of Santorini and the pancreatic duct were re-approximated to the duodenal wall to prevent acute pancreatitis caused by scarring and stenosis of the duct orifice. Histological findings were consistent with well-differentiated adenocarcinoma limited to the minor duodenal papilla, without infiltration of the duodenal wall submucosa, and confirmed complete resection. The patient had an uneventful postoperative course and has remained asymptomatic, without evidence of tumor recurrence or stenosis of the pancreatic duct orifice, for 4 years.

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Key words: adenocarcinoma, minor papilla, transduodenal minor papillectomy

Introduction

Tumors of the minor duodenal papilla are rare, although adenoma¹, adenomyoma², carcinoid^{3,4}, and somatostatinoma^{5,6} have been reported. Malignancy affecting the minor papilla in the region of the accessory pancreatic duct is also rare, with only 7 reported cases^{7–13}. All 7 patients underwent pancreaticoduodenectomy for primary adenocarcinomas of the minor duodenal papilla. The

reported frequencies of malignancy in adenomas of the papillae ranges from 26% to 65%^{14–16}. Thus, adenomas of the major or minor papilla are thought to exhibit the same adenoma-carcinoma sequence as do adenomas of the colon. Although there is a consensus that papillary adenomas greater than 1.0 cm in size should be resected, the procedure of choice remains controversial. Traditionally, these tumors have been treated with pancreaticoduodenectomy. Early ampullary cancer is defined as a tumor limited to the mucosa of the

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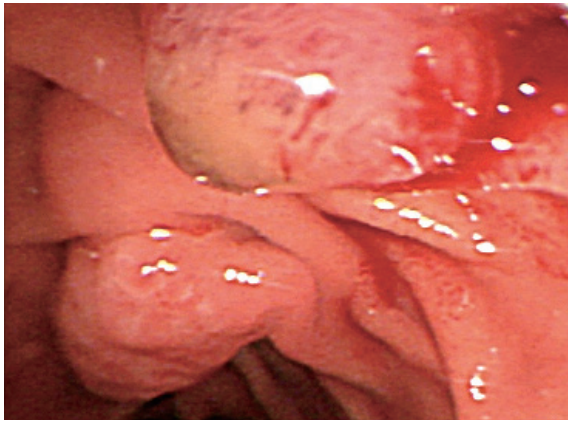


Fig. 1 Duodenoscopy with a side-view endoscope showed that the tumor had originated in the minor duodenal papilla. The major papilla was to the anal side of the minor papilla.

ampulla (depth of cancer invasion: m) or the sphincter of Oddi (depth of cancer invasion: od) regardless of the presence or absence of lymph-node metastasis¹⁷, which corresponds to pT1 of the World Health Organization classification¹⁸. We herein report a case of adenocarcinoma of the minor duodenal papilla treated with transduodenal excision of the minor papilla.

Case Report

A 64-year-old woman was admitted to our hospital for treatment of an asymptomatic duodenal tumor. The patient's general practitioner had found an irregular elevated mass in the second part of the duodenum on routine screening upper gastrointestinal endoscopy. The medical history was significant only for hypertension. The familial history included her mother who died of gallbladder cancer and a sister with rectal cancer. The patient was in the process of quitting habitual tobacco smoking and alcohol consumption.

There were no abnormal findings on physical examination. Results of laboratory studies, including the hematological profile, renal function, pancreatic enzymes, liver enzymes, electrolytes, and tumor markers (CA19-9 and CEA), were all within normal limits. Abdominal computed tomography (CT) failed to demonstrate the primary tumor in the duodenum and did not reveal distant metastasis, lymph node

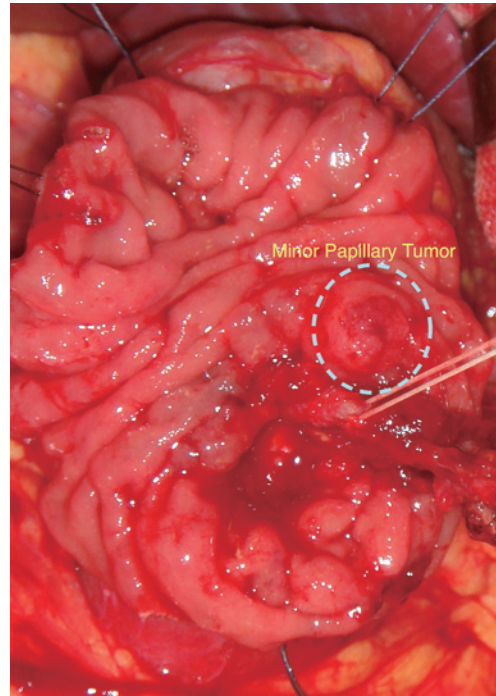


Fig. 2 The tumor in the minor papilla. Transduodenal excision of the minor papilla was performed.

metastasis, or peritoneal dissemination. Magnetic resonance cholangiopancreatography showed the pancreatic duct more clearly than did CT. Endoscopic ultrasonography failed to show any masses in the minor duodenal papilla. Screening upper gastrointestinal endoscopy revealed a 15-mm sessile tumor in the descending duodenum proximal to the major papilla (**Fig. 1**). Magnifying endoscopic images showed both the major and minor papillae to be enlarged with normal surfaces. A villous polypoid tumor, approximately 24 mm in diameter, was observed at the major duodenal papilla. Biopsy results of these tissues were consistent with tubular adenoma with moderate epithelial atypia.

Transduodenal excision of the minor papilla including the tumor was performed, and the duct of Santorini was re-approximated to the duodenal wall with 4-0 absorbable sutures (**Fig. 2**). The same procedure was performed for the tumor of the major papilla. The common bile duct and the pancreatic duct were sutured and re-approximated to the duodenal wall with 4-0 absorbable sutures (**Fig. 3**). No prophylactic pancreatic stents were placed.

Histopathological examination of the resected

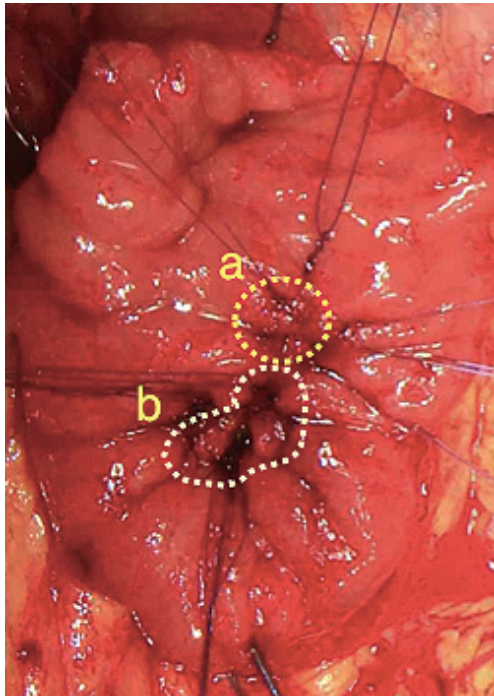


Fig. 3 (a) The duct of Santorini is re-approximated to the duodenal wall with 4-0 absorbable suture for the Santoriniplasty. (b) The common bile duct and the pancreatic duct were sutured and re-approximated to the duodenal wall with 4-0 absorbable sutures.

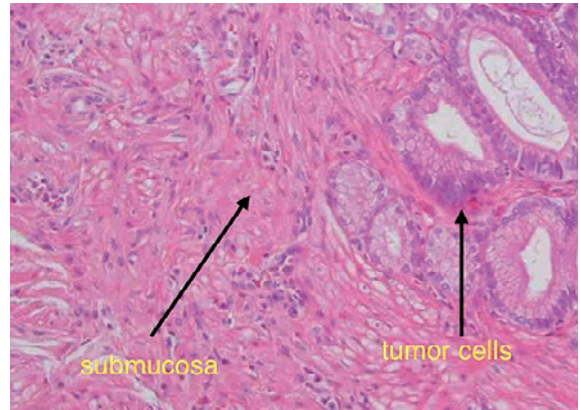


Fig. 4 Microscopic examination revealed a well-differentiated adenocarcinoma within the minor duodenal papilla, without infiltration of the submucosa of the duodenal wall, and confirmed complete resection.

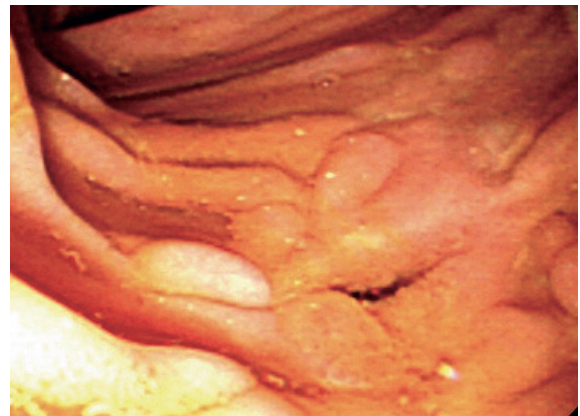


Fig. 5 Endoscopic view of the orifice of the pancreatic duct and the common bile duct 4 years after the operation. There was no evidence of tumor recurrence, and the patency of the duct of Santorini was maintained.

specimen revealed a well-differentiated adenocarcinoma within the minor duodenal papilla, without infiltration to the submucosa of the duodenal wall, and confirmed complete resection (Fig. 4). According to the World Health Organization classification¹⁸, the histological stage of the tumor was pT1. The pathological diagnosis of the tumor of the major papilla was adenoma with moderate atypia. Additional surgical treatment was discussed with the patient and her family, but they refused. The patient had an uneventful postoperative recovery and was discharged on the 10th postoperative day.

Postoperative adjuvant therapy was not performed because of the early tumor stage. In the 4 years since the surgery, the patient has been asymptomatic without evidence of tumor recurrence or stenosis of the pancreatic and common bile duct orifice on endoscopic examination (Fig. 5).

Discussion

Tumors in the region of the minor papilla are rare, and most of the reported cases are benign tumors, such as carcinoids³⁴ and somatostatinoma⁵⁶. In fact, the incidence of adenocarcinoma of the minor duodenal papilla, especially at an early stage, might be low¹². A possible reason for the small number of reported cases is that most tumors in this area seldom produce symptoms in the early stage and are identified only at an advanced stage. The lack of characteristic symptoms makes tumors of the minor

Table 1 Documented cases of adenocarcinoma of the minor duodenal papilla

Case no.	First author	Sex	Age (years)	Chief complaint	Size (mm)	Histologic diagnosis	Invasion of pancreas	Treatment	Cancer of major papilla
1	Yamao ⁷⁾	M	82	Transient epigastric pain	25 × 20	Mod	+	PpPD	–
2	Kajiwara ⁸⁾	M	60	Transient gastric pain	50 × 30	Well	+	SSpPD	–
3	Wakatsuki ⁹⁾	M	70	no	11 × 8	Well	–	PpPD	–
4	Parthasarathy ¹⁰⁾	F	60	Fever and jaundice	15 × 12	Mod	+	PD	+
5	Matheus ¹¹⁾	F	50	Abdominal pain and jaundice	10	Mod	–	PpPD	+
6	Okuma ¹³⁾	M	76	no	12 × 10	Well	–	PpPD	–
7	Takami ¹²⁾	M	81	no	20 × 15	Pap + Well + Mod +	+	SSpPD	–
8	Present case	F	64	no	15	Well	–	TP	–

Mod, moderately differentiated adenocarcinoma; Well, well-differentiated adenocarcinoma; Pap, papillary adenocarcinoma; PpPD, pylorus-preserving pancreatoduodenectomy; SSpPD, subtotal stomach-preserving pancreatoduodenectomy; PD, pancreatoduodenectomy; TP, transduodenal papillectomy

duodenal papilla difficult to diagnose.

Only 7 adenocarcinomas of the minor papilla have been reported previously^{7–13}. The present case was also asymptomatic, and the tumor, 1.5-cm proximal to the major duodenal papilla, had been identified incidentally on screening upper gastrointestinal endoscopy. Two of the previous 7 cases (cases 4 and 5) involved synchronous carcinomas of the major and minor duodenal papillae^{10,11} (**Table 1**). Neither of these cases involved invasion of the minor papilla. The tumors of the major and minor papillae were not related. There was no pancreaticobiliary maljunction, thus they had examined with magnetic resonance cholangiopancreatography image or endoscopic retrograde cholangiopancreatography for anomalous junction of the pancreaticobiliary duct. Of the 7 previously reported tumors of the minor duodenal papilla, 3 were asymptomatic^{9,12,13}, 2 were associated with jaundice^{10,11}, and 2 were associated with transient epigastralgia^{7,8} (**Table 1**). Because none of these cases had symptoms specifically associated with tumors of the minor duodenal papilla, careful observation of the papilla during routine endoscopic examinations of the upper gastrointestinal tract is essential.

In the case of the major duodenal papilla, the reported rates of malignancy involving duodenal or ampullary tumors range from 35% to 60%^{19,20}. While

the region of the ampulla is easily accessible for endoscopic biopsy, the high rate of false-negative results for carcinoma are high, from 25% to 60%^{19,20}. Differentiating benign from malignant tumors on the basis of size or external appearance is difficult. Therefore, the tumor was completely resected in our patient because of the potentially serious problems of endoscopic biopsy and external appearance described above. We believe that adenomas of the major or the minor papilla undergo the same adenoma-carcinoma sequence as adenomas of the colon. However, a consensus has reportedly been reached that papillary adenomas greater than 1.0 cm in size should be resected²¹.

Some recently published series found endoscopic snare excision of papillary adenoma to result in favorable outcomes in terms of both safety and long-term efficacy. However, a few complications and minimal residual tumors after endoscopic resection have also been observed^{21–23}. Some cases included acute pancreatitis due to stenosis of the pancreatic duct orifice as a late complication^{21–23}. In the present case, the tumors were located at both the major and minor papillae. We were thus concerned that pancreatitis might develop in association with complete stenosis of the pancreatic duct orifice. Therefore, we decided to perform transduodenal excision of the minor papilla, followed by

Santoriniplasty.

In all 7 previously reported cases, pancreaticoduodenectomy was performed to treat primary adenocarcinomas of the minor duodenal papilla⁷⁻¹³ (**Table 1**). Preoperative examination in the present case showed the tumor to be malignant, and we thus considered pancreaticoduodenectomy including laparoscopic surgery²⁴ to be necessary. Our patient showed no signs of malignancy preoperatively. En bloc resection was performed in case malignant cells were present in the surrounding tissues. Although Ohta and Takazaki²⁵ have reported that the risk of nodal metastasis is low when a carcinoma of the papilla of Vater cancer does not invade beyond the sphincter of Oddi, some researchers have reported higher rates of lymphatic permeation (22%) and nodal metastasis (10%, 11%) in cases with invasion of the sphincter of Oddi^{26,27}. In the present case, the tumor had invaded beyond the basement membrane into the sphincter of Oddi. According to the General Rules for Surgical and Pathological Studies on Cancer of the Biliary Tract by the Japanese Society of Biliary Surgery, carcinoma of the papilla of Vater confined to the mucosa and contiguous with the epithelium of the glands in the sphincter of Oddi is defined as mucosal cancer¹⁷. There is a possibility that cancer of the minor papilla of Vater carries no risk of metastasis that invasion does not reach the mucosa. The present case has also been asymptomatic with neither macroscopic nor histological recurrence for 48 months since surgery. Consequently, the orifice of the pancreatic duct has maintained an adequate size for 4 years (**Fig. 5**). Our experience in this case suggests that transduodenal excision is a feasible alternative treatment for papillary tumors.

Conflict of interest statement: None of the authors has any conflicts of interest.

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