

CASE REPORT

## Endoscopic papillectomy of minor papillar adenoma associated with pancreas divisum

Akira Kanamori, Takashi Kumada, Seiki Kiriya, Yasuhiro Sone, Makoto Tanikawa, Yasuhiro Hisanaga, Hidenori Toyoda, Hiroki Kawashima, Akihiro Itoh, Yoshiki Hirooka, Hidemi Goto

Akira Kanamori, Takashi Kumada, Seiki Kiriya, Yasuhiro Sone, Makoto Tanikawa, Yasuhiro Hisanaga, Hidenori Toyoda, Department of Gastroenterology, Ogaki Municipal Hospital, Ogaki 503-8502, Japan

Hiroki Kawashima, Akihiro Itoh, Hidemi Goto, Department of Gastroenterology, Nagoya University Graduate School of Medicine, Nagoya 466-8560, Japan

Yoshiki Hirooka, Hidemi Goto, Department of Endoscopy, Nagoya University Hospital, Nagoya 466-8560, Japan

Author contributions: All authors contributed equally to this work; Akira Kanamori wrote the paper.

Correspondence to: Akira Kanamori, Department of Gastroenterology, 4-86 minaminokawa-cho, Ogaki, Gifu, 503-8502, Japan. [tkumada@he.mirai.ne.jp](mailto:tkumada@he.mirai.ne.jp)

Telephone: +81-584-813341 Fax: +81-584-755715

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### Abstract

Tumors of the minor papilla of the duodenum are quite rare. We successfully and safely treated an 18-mm adenoma of the minor papilla associated with pancreas divisum using endoscopic papillectomy. A 64-year-old man was admitted to our hospital for treatment of an asymptomatic mass in the minor papilla detected by upper gastrointestinal endoscopy. Endoscopic analysis showed an 18-mm, whitish, sessile mass, located in the duodenum proximal to a normal-appearing major papilla. Endoscopic retrograde pancreatography did not reveal the pancreatic duct. Magnetic resonance cholangiopancreatography showed a lack of the ventral pancreatic duct. We suspected this case was associated with pancreatic divisum; therefore, we performed endoscopic papillectomy of the minor papilla tumor. Subsequently, endoscopic pancreatic stent placement in the minor papilla was done to prevent drainage disturbance. The patient has been asymptomatic without recurrence of tumor or stenosis of the Santorini orifice upon endoscopic examination for the past 2 years.

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**Key words:** Endoscopic papillectomy; Minor papillar adenoma; Pancreas divisum; Endoscopic pancreatic stent; Endoscopic retrograde pancreatography

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### INTRODUCTION

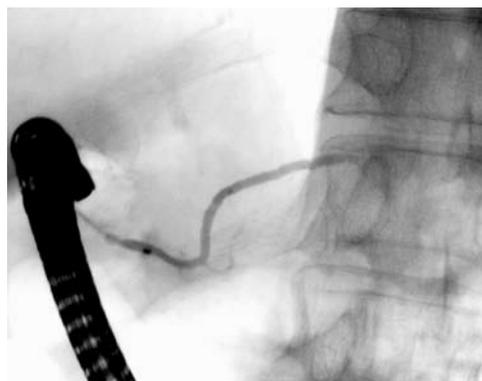
Tumors arising in the region of the major duodenal papilla account for 5% of gastrointestinal (GI) neoplasms and 36% of resectable pancreaticoduodenal tumors<sup>[1]</sup>. Adenoma is a particularly common finding in patients with familial adenomatous polyposis (FAP). However, currently, adenomas that involve the papilla have been recognized increasingly often, even in the absence of FAP. With the development of endoscopic tools, the safety and the efficacy of endoscopic papillectomy has improved, and indications for endoscopic papillectomy have recently been expanded<sup>[2-5]</sup>. Recently, endoscopic papillectomy has been accepted as a viable alternative therapy to surgery in sporadic ampullary adenoma and has yielded high success and low recurrence rates<sup>[5,6]</sup>. However adenoma of the minor papilla has been reported in only a few cases<sup>[7-9]</sup>. We report a case of endoscopic treatment of sporadic adenoma of the minor papilla associated with pancreas divisum.

### CASE REPORT

A 64 year-old man was admitted to Ogaki Municipal Hospital for evaluation of an asymptomatic duodenal tumor that was found incidentally by X-ray examination of the stomach during a periodic health examination. The patient's medical history was otherwise unremarkable, and he had no family history of FAP. The laboratory findings were within normal limits, including tumor markers. Endoscopic analysis showed an 18-mm, whitish, elevated, slightly rough-surfaced mass, located in the descending duodenum proximal to



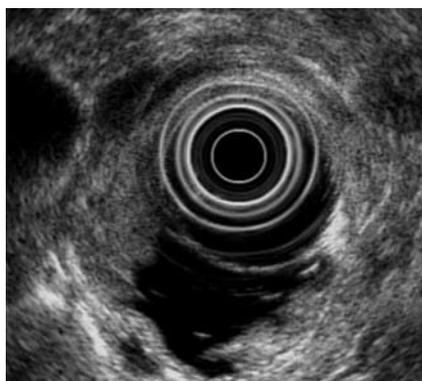
**Figure 1** Endoscopy showing an 18-mm, whitish, elevated, slightly rough-surfaced mass, located proximal to the major papilla.



**Figure 4** ERP was immediately performed *via* the minor papilla and it showed that the entire dorsal pancreatic ductal system was without communication with the ventral pancreatic duct.



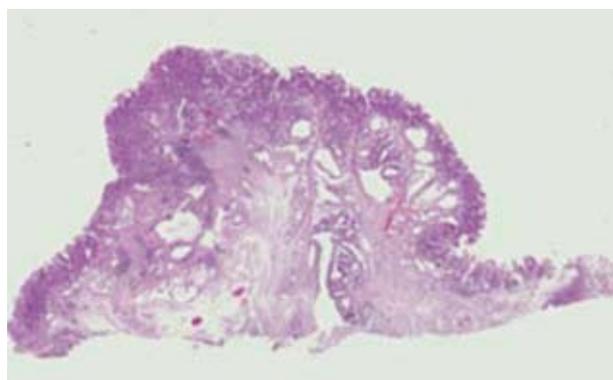
**Figure 2** Hypotonic duodenography demonstrating a mass situated 15 mm proximal to the major papilla, which was raised highly from the duodenum.



**Figure 3** EUS detected an 18 mm × 12 mm homogeneous, hypoechoic mass in the submucosal layer.



**Figure 5** A pancreatic 5Fr stent was placed immediately after endoscopic papillectomy and coagulated the margin of the minor papilla tumor.



**Figure 6** Histopathological findings of the specimen showed tubular adenoma and the margin of the tumor was negative; however, a slight infiltration of the pancreatic duct system was revealed (HE stain, × 4).

a normal-appearing major papilla (Figure 1). Histological examination of forceps biopsy specimens from the mass revealed a tubular adenoma with moderate epithelial atypia. Hypotonic duodenography (double contrast radiographic study) demonstrated a mass situated 15 mm proximal to the major papilla, which was raised highly from the duodenum (Figure 2). Endoscopic retrograde pancreatography (ERP) (JF-230; Olympus, Tokyo, Japan) did not reveal the pancreatic duct upon initial examination.

Magnetic resonance cholangiopancreatography (MRCP) showed the entire dorsal pancreatic duct and the lack of a ventral pancreatic duct. We suspected this case was associated with pancreas divisum and that the tumor had arisen from the minor duodenal papilla. Endoscopic ultrasonography (EUS) (GF-UM240; Olympus) detected an 18 mm × 12 mm homogeneous,

hypoechoic mass in the submucosal layer (Figure 3). We further determined that the tumor was not invasive to the muscle layer. Endoscopic papillectomy of the minor papilla was performed after obtaining appropriate written informed consent. Submucosal injection of hypertonic saline-epinephrine (HSE) was carried out, and subsequently, snare excision was performed with polypectomy snare forceps. Following this, ERP was immediately performed *via* the minor papilla and it showed that the entire dorsal pancreatic ductal system was without communication with the ventral pancreatic duct (Figure 4). We placed a prophylactic pancreatic 5Fr stent (Figure 5) and endoscopic papillectomy was

performed successfully without any procedure-related complications. The resected specimen showed tubular adenoma with moderate epithelial atypia in the mucosal layer. However, the margin of the tumor was negative, with a slight infiltration of the pancreatic duct system (Figure 6). One week later, duodenoscopy was performed and no evidence of remaining tumor was seen, and the pancreatic stent was withdrawn. For the last 2 years, the patient has been asymptomatic without evidence of tumor recurrence or stenosis of the pancreatic duct orifice, based on endoscopic examinations performed every 6 mo.

## DISCUSSION

Endoscopic papillectomy of minor papilla tumors has been reported in only a few cases. Adenoma of the minor papilla associated with pancreas divisum is particularly rare and has been reported previously only once by Nakamura *et al*<sup>[8]</sup>. We think that duodenal and periampullary tumors occur in the general population, although patients with FAP invariably develop duodenal adenomas and have a risk of papillary carcinoma<sup>[10]</sup>. However, with the development of endoscopic tools and techniques, papillectomy has been accepted as a safe and feasible treatment for adenoma of the major papilla. It is important to diagnose tumor ductal infiltration correctly to determine endoscopic respectability by using intraductal ultrasonography (IDUS)<sup>[11]</sup>. In the present case, because ERP *via* the major and minor papilla was unsuccessful before the treatment of minor papillary tumors, we performed EUS before treatment and diagnosed the tumor as non-invasive to the muscle layer. EUS is a highly accurate and non-invasive modality for staging ampullary neoplasms and for evaluating ductal involvement by a tumor<sup>[12]</sup>. However, it is also essential to accurately diagnose tumors, adenoma or early cancer as not infiltrating Oddi's muscle layer. Therefore, we think it necessary to undertake IDUS before treatment of minor tumors of the papilla as frequently as possible.

In the present case, we injected HSE into the submucosal layer upon endoscopic papillectomy to reduce the risk of perforation. It is uncertain whether submucosal injections reduce the risk of perforation upon endoscopic papillectomy of a tumor of the major papilla. We think it safer to inject HSE into the submucosal layer upon endoscopic papillectomy of the tumor of the minor papilla. Many authors have reported the efficacy of pancreatic stents for decreasing both post-procedure pancreatitis and stenosis<sup>[13,14]</sup>. In the present case, the minor papilla drained all of the pancreatic juice flow from the dorsal pancreas. Therefore, we considered the placement of a pancreatic stent to be essential after endoscopic papillectomy of the minor papillary tumor.

We performed follow-up duodenoscopy and computed tomography at 3, 6, 12 and 18 mo later, and there was no evidence of tumor recurrence or

stenosis of the orifice of the Santorini duct for more than 2 years. Some authors have reported that long-standing pancreatic duct obstructions caused by relative stenosis of the minor duodenal papilla might be a factor promoting oncogenesis<sup>[15]</sup>. We will be performing a follow-up study of recurrence of minor papillary tumors and careful surveillance of the duodenum and pancreaticobiliary system.

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