
Carcinoma Arising from Brunner's Gland in the Duodenum after 17 Years of Observation – A Case Report and Literature Review

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Key Words

Brunner's gland · Adenocarcinoma · Duodenal cancer

Abstract

A 60-year-old man presented with melena and hematemesis in 1984. Esophagogastroduodenoscopy (EGD) detected a small protruding lesion in the duodenal bulb, which was diagnosed as Brunner's adenoma. No significant change was detected in subsequent annual EGD and biopsies for 10 years, after which the patient was not observed for 7 years. The patient presented with melena again in 2001. The lesion had changed shape to become a 10 mm sessile tumor with a central depression, and following a biopsy was diagnosed as an adenocarcinoma. The patient underwent partial resection of the duodenum. Histopathological assessment showed acidophilic cells with swollen nuclei, and clear cells forming a tubular or papillary tubule in the mucosal lamina propria and submucosal layer. The tumor cells stained positive for lysozyme, indicating that they arose from Brunner's gland. The patient showed no sign of recurrence and was disease-free for more than 34 months after surgery. The patient died of pneumonia. This is an extremely rare case of primary duodenal carcinoma arising from Brunner's gland in a patient observed for 17 years.

Introduction

The increased use of endoscopies in recent years has led to an increase in the identification of duodenal lesions; however, primary duodenal carcinomas remain comparatively rare and account for only 0.3% of digestive organ carcinomas [1–3]. Furthermore, tumors arising from Brunner's gland are extremely rare [4–23], and only

two reports describe the preoperative observation period [11, 21]. Here we report on an extremely rare case of primary duodenal carcinoma arising from Brunner's gland in a patient who had been under observation for 17 years.

Case Report

A 60-year-old man presented with melena and hematemesis in 1984. Esophagogastroduodenoscopy (EGD) detected a small protruding lesion in the duodenal bulb, which was diagnosed as Brunner's adenoma. Subsequent annual EGD and biopsies showed no significant changes for 10 years, after which the patient was lost to follow-up for 7 years. He again presented with melena in 2001. The lesion had changed shape to become a 10 mm sessile tumor with a central depression according to EGD (fig. 1) and hypotonic duodenography (fig. 2), and following a biopsy was diagnosed as adenocarcinoma. Laboratory test results, including those from tumor marker (CEA and CA19-9) assays, were within normal limits. The patient underwent partial resection of the duodenum.

Histopathology assessment found acidophilic cells with swollen nuclei, and clear cells forming a tubular or papillary tubule in the mucosal lamina propria and submucosal layer (fig. 3a, b). Immunohistological staining showed the tumor to be negative for chromogranin-A, p-53, lipase and amylase. Immunohistochemical staining showed the tumor cells to be positive for MIB-1 (fig. 3c) and lysozyme (fig. 3d), indicating that they arose from Brunner's gland.

The patient showed no sign of recurrence in abdominal computed tomography and tumor markers and was disease-free for more than 34 months after surgery. He subsequently died of pneumonia.

Discussion

Duodenal carcinoma may arise in different types of cells of the duodenal mucosa. It has been proposed that they arise as either de novo lesions, or from adenomas or aberrations of the pancreas or gastric mucosa [24, 25]. Carcinomas arising from Brunner's gland are very rare, and only 21 such cases have been reported in the literature, the first of which was by Shorrock et al. in 1986 [4]. Immunohistochemical examination is essential for determining the origin of carcinomas arising from Brunner's gland. In the present case, a Brunner's gland adenocarcinoma was indicated by positive MIB-1 and lysozyme staining, the absence of a surrounding normal Brunner's gland, and negative staining for chromogranin A, p53, lipase and amylase.

In 2002, Akino et al. summarized 16 cases of carcinoma arising from Brunner's gland [3, 4, 9, 11, 14, 19, 26, 27], and five further cases were later reported [20–23]. Our analysis of these 20 cases shows they involved 15 men and 5 women, with a mean age of 67.4 years (range 39–85 years) [4–23] (table 1). Eight tumors were located in the first portion of the duodenum and twelve in the second portion. In terms of shape, five tumors mimicked submucosal tumors, seven were sessile, four were type 2 carcinomas, and four were polypoid lesions. The mean tumor diameter was 25.6 mm (range 7–70 mm). Four tumors were limited to the mucosal layer, nine showed submucosal invasion, and six were advanced carcinomas. The variety in the tumor forms reflects that Brunner's gland exists in the deep lamina propria of the mucosa or in a submucosal layer [28]. All tumors were diagnosed as highly differentiated adenocarcinomas. Ten cases underwent limited resection, comprising six partial resections of the duodenum, two endoscopic mucosal resections, and two polypectomies. Those ten resection cases showed relatively good outcomes over a mean observation period of 21.6 months (range 3–45 months).

The present case was unique in that it involved an adenocarcinoma developing after 17 years of observation. Other cases have been reported after 5 years and 3.3 years of observation, confirming the slow growth of this tumor type [11, 21]. De novo malignant

degeneration of Brunner's gland has rarely been described [27, 29]. Given the nature of Brunner's gland adenocarcinoma, it should be removed if its shape or size changes significantly.

Conclusions

We report on an extremely rare case of a primary duodenal carcinoma arising from Brunner's gland in a patient under observation for 17 years. It is concluded that Brunner's gland adenomas should be followed up sequentially, and be removed if their shape or size changes significantly.

Table 1. Published cases of duodenal cancer arising from Brunner's gland (1986–2007)

No.	Author	Year	Age	Gender	Location	Macroscopic appearance	Maximum size (mm)	Depth of invasion	Histological type	Preoperative observation time (years)	Operation	Outcome (months)
1	Shorrock [4]	1986	51	M	2nd	polypoid	unknown	sm	adenocarcinoma	–	PD	A (18)
2	Ohmachi [5]	1989	64	M	1st	sessile	7	sm	pap	–	DG	A (9)
3	Kubota [6]	1990	70	M	1st	type 2	70	ss	pap - tub	–	PD, TG, RHC	unknown
4	Fukuda [7]	1990	72	M	1st	sessile	15	sm	adenocarcinoma	–	PR	unknown
5	Sasaki [8]	1991	66	F	1st	type 2	50	si (panc)	muc	–	PD	A (9)
6	Miyamoto [9]	1991	45	F	1st	sessile	19	m	tub1	–	PR	unknown
7	Sakai [10]	1992	39	F	2nd	type 2	40	ss	tub1	–	PD	A (18)
8	Itsuno [11]	1993	75	M	1st	sessile	14	unknown	pap	3.3	–	D (3)
9	Komatsu [13]	1994	71	M	2nd	SMT	33	a1	tub1	–	PD	D (45)
10	Kawamoto [14]	1994	69	M	1st	SMT	20	sm	pap	–	PR	A (36)
11	Suzuki [15]	1995	77	M	2nd	polypoid	15	m	tub1	–	polypectomy	unknown
12	Arai [16]	1998	55	F	2nd	SMT	17	sm	tub1	–	PR	A (21)
13	Shinohara [17]	2000	71	M	2nd	polypoid	24	sm	tub1	–	PR	A (6)
14	Kobayashi [18]	2001	75	M	2nd	SMT	12	sm	adenocarcinoma	–	PR	unknown
15	Akino [19]	2002	82	M	2nd	SMT	10	m	adenocarcinoma	–	EMR	A (36)
16	Kushima [20]	2002	85	F	2nd	polypoid	20	m	adenocarcinoma	–	polypectomy	A (28)
17	Miyamoto [21]	2003	57	M	2nd	type 2	15	mp	sig	5	PD	A (24)
18	Sakurai [22]	2005	73	M	2nd	sessile	13	sm	adenocarcinoma	–	unknown	A (20)
19	Sakurai [22]	2005	73	M	1st	sessile	78	ss	adenocarcinoma	–	unknown	D (27)
20	Kimura [23]	2007	77	M	2nd	sessile	15	sm	adenocarcinoma	–	EMR	A (unknown)
21	Our case	2007	77	M	1st	sessile	10	sm	adenocarcinoma	17	PR	D (34)

Macroscopic appearance: SMT = submucosal tumor-like. Depth of invasion: m = mucosa; sm = submucosa; mp = muscularis propria; ss = subserosa; si = invade adjacent structure; panc = pancreas. Histological type: pap = papillary adenocarcinoma; tub = tubular adenocarcinoma; tub1 = tubular adenocarcinoma well differentiated type; muc = mucinous adenocarcinoma; sig = signet-ring cell carcinoma. Operation: PD = pancreatoduodenectomy; DG = distal gastrectomy; TG = total gastrectomy; RHC = right hemicolectomy; PR = partial resection; EMR = endoscopic mucosal resection. Outcome: A = alive; D = dead.

Fig. 1. Esophagogastroduodenoscopy showing a 10 mm sessile tumor with a central depression in the duodenal bulb.



Fig. 2. Hypotonic duodenography showing a contrast media-positive elevated lesion in the center of the duodenal bulb.

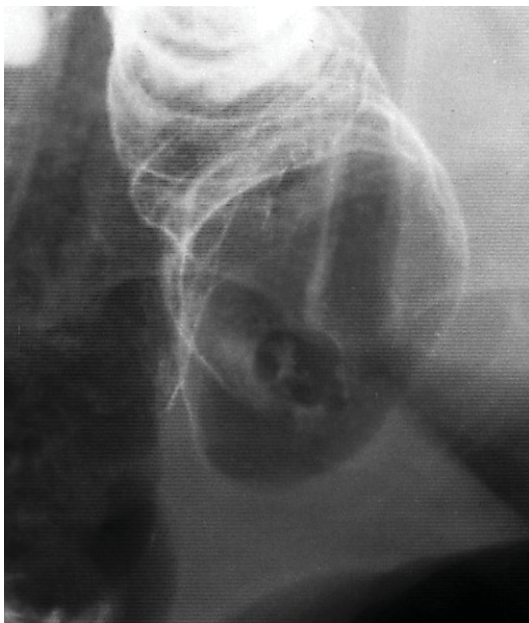
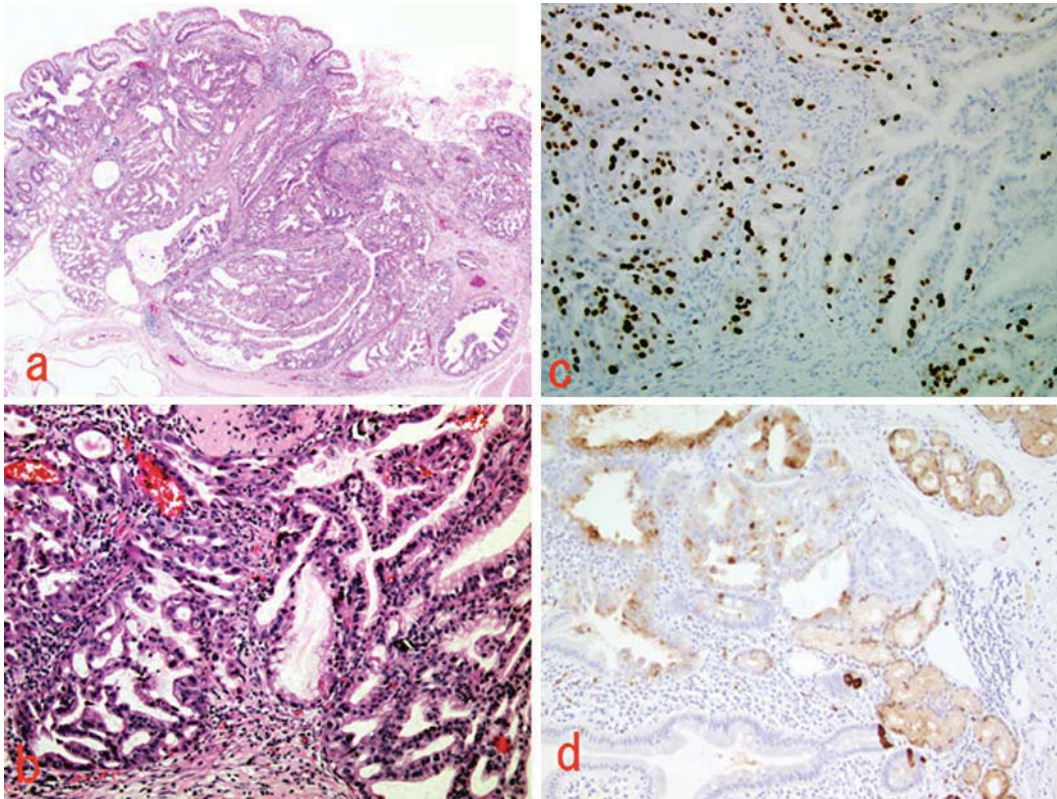


Fig. 3. Microscopic findings of the tumor. **a** Gross appearance following hematoxylin and eosin staining ($\times 2$). **b** Adenocarcinoma in an adenoma (hematoxylin and eosin stain, $\times 10$). **c** MIB-1-positive neoplastic cells ($\times 10$). **d** Lysozyme-positive neoplastic cells ($\times 10$).



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