Case report

Early gastric cancer located just above Dieulafoy's ulcer, with massive bleeding

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Abstract

In 2003, a 69-year-old man visited our emergency department because of hematemesis and anemia. Emergency gastroscopy revealed massive bleeding from Dieulafoy's ulcer in the upper body of the stomach. The arterial bleeding was successfully controlled by endoscopic clipping. Blood transfusion and a proton-pump inhibitor were administered and his condition recovered smoothly. Two weeks after the treatment, type IIa early gastric cancer was detected at the previous bleeding point by follow-up endoscopy. He underwent distal gastrectomy with systematic lymph node dissection (D2), and he had no sign of recurrence until 2005. Histopathological examination revealed an early gastric cancer with submucosal invasion located just above the Dieulafoy's disease. The characteristic finding of Dieulafoy's disease was an enlarged and tortuous artery arising from the subserosa, penetrating the muscle layer, and spreading in the submucosa. Abnormal Dieulafoy's artery coexisting with gastric cancer has been reported in only 17 cases until now. Our clinical and pathological findings led us to the following speculation on the pathogenesis in our patient. Repeated regeneration of the mucosal membrane would have been caused by circulatory disturbance in Dieulafoy's vessels. This regeneration and mucosal dysplasia may have been a factor in promoting the gastric cancer. In the previously reported cases of the coexistence of abnormal Dieulafoy's artery and gastric cancer, the initial gastroscopic examination rarely diagnosed the gastric cancer. Thus, follow-up gastroscopy is essential, so as not to miss such coexisting diseases.

Key words Gastric cancer · Dieulafoy's ulcer · Massive bleeding · Endoscopic hemostasis

Introduction

Dieulafoy's disease is an important cause of gastrointestinal (GI) hemorrhage, accounting for up to 5% of acute

GI bleeding. Although first reported by Gallard in 1896, the lesion was more accurately described by Dieulafoy in 1897. The histopathological finding is characteristic; an enlarged and tortuous artery lies in close proximity to the mucosal surface, likely as a congenital anomaly. The most frequent bleeding point is the gastric fundus or body. Hemorrhage is often massive and may be life-threatening [1].

Dieulafoy's disease coexisting with gastric cancer is very rare, with 17 cases having been reported up to now [2–5] since Sasaki first reported a case in 1982. We report the successful treatment of a patient with an early gastric cancer located just above a Dieulafoy's ulcer.

Case report

A 69-old-man visited the emergency department of Horie General Hospital, Japan, with complaints of epigastric discomfort, repeated hematemesis, and progressive anemia, on October 30, 2003. He had a history of duodenal ulcer at the age of 25. Since 2000, he had been treated with a calcium antagonist for hypertension and with sulfonyl urea for diabetes mellitus.

Emergency gastroscopy revealed massive coagula and spurting bleeding from the posterior wall of the upper third of the stomach. The bleeding was successfully stopped with an endoscopic hemoclip, using a clear attachment. Blood transfusion and a proton-pump inhibitor were administered and he recovered smoothly. He was discharged 20 days after the treatment. Follow-up gastroscopy was performed 2 weeks and 6 weeks after the treatment. Two weeks after the treatment, we found a slightly elevated mucosal cancer, type IIa [6], at the previous bleeding point (Fig. 1). Well-differentiated adenocarcinoma was also confirmed by endoscopic biopsy. Double-contrast radiography showed irregular mucosal convergence at the site.

Fig. 1a,b. Follow-up gastroscopy. **a** Type IIa early gastric cancer in upper posterior wall; **b** indigocarmine dye stain

Fig. 2. Resected specimen, showing type IIa cancer with shallow depression at the center (arrows, marking clips for operation)

The patient was admitted again, and underwent distal gastrectomy with systematic lymph node dissection (D2) [6]. He had a good postoperative recovery and showed no sign of recurrence until February 2, 2005. A type IIa early gastric cancer with a small central shallow depression was confirmed in the resected specimen (Fig. 2). The size was 50 × 40 mm. Microscopic examination revealed well-differentiated adenocarcinoma with slight invasion of the submucosal layer (sm). There was no metastasis in the removed lymph nodes. Furthermore, an enlarged and tortuous artery was found arising from the serosa, penetrating through the muscle layer, and spreading in the submucosa (Fig. 3a,b). This artery,

which had ordinary elastic lamina in the wall, was obstructed by thickening of the intima around the ulcer scar, caused by the hemostasis. Neither arteriovenous malformation nor aneurysm was present (Fig. 3c, d, e).

Discussion

Dieulafoy's ulcer causes massive bleeding from a shallow ulceration of the GI tract. Recently it was discovered that these ulcers could occur at any site of the GI tract; stomach, duodenum, colon, rectum, etc. However, two-thirds of them were located in the stomach [1]. Similarly, the bleeding point was found in the stomach — on the posterior wall of the gastric body — in our patient.

Currently, the bleeding is controlled by endoscopic hemostasis. Treatments are classified into three methods [7,8]: (1) a heater-probe method; (2) the submucosal injection of ethanol, hypertonic saline epinephrine solution, or a sclerosing agent such as ethoxysclerol; and (3) a mechanical hemoclip. We preferred to use a hemoclip hemostasis because of its reliability and minimal tissue damage, and we succeeded in stopping the bleeding.

We could not diagnose the early gastric cancer at the bleeding point on the initial gastroscopy, because the tumor elevation was slight and it was covered with blood coagula. We were able to detect the cancer just above the ulcer on the follow-up gastroscopy. This is an important message: that follow-up endoscopy is essential not to miss coexisting disease. The cancer was successfully treated by distal gastrectomy with systematic lymph node dissection. In addition, we found neither an

Fig. 3a-f. Microscopic examinations. **a** Type IIa early gastric cancer and abnormal artery penetrating through the muscle layer. **b** Cancer cell infiltration (arrows) into submucosa. **c** Obstructed artery (arrow) near the ulcer scar. **d** Obstructed artery with thickened intima and small recanalization. **e**

Elastic lamina shown in Dieulafoy's artery (spreading in the submucosa). **f** Coexistence of early gastric cancer and Dieulafoy's disease (diagram). **a** H&E, $\times 4$; **b** H&E, $\times 10$; **c** H&E, $\times 4$; **d** H&E, $\times 25$; **e** elastic van Gieson, $\times 4$

Table 1. Reported cases of gastric cancer found by Dieulafoy's bleeding

No.	Author	Year	Age (years)	Sex	Location	Ca. position	Size (mm)	Type	Histology	Depth	Diagnosis	Endo. treatment
1	Sasaki	1982	66	M	Unknown	On Dieulafoy	Unknown	Ilc + III	Unknown	sm	Benign	(-)
2	Maeba	1986	63	M	C. Ant.	On Dieulafoy	20 × 20	Ilc	tub	sm	Benign	(-)
3	Fujimori	1988	47	F	M. Less.	On Dieulafoy	Unknown	Ilc	tub 2	m	Benign	(-)
4	Fujimori	1988	42	M	C. Ant.	On Dieulafoy	Unknown	Ilc	tub 2	sm	Benign	(-)
5	Kawamura	1991	32	F	C. Ant.	On Dieulafoy	16 × 12	Ilc + III	tub 2	m	Ilc	(-)
6	Natsugoe	1991	65	M	C. Post.	On Dieulafoy	22 × 17	Ilc	tub 1	sm	Ilc	Ethanol + HSE
7	Taniguchi	1992	48	M	M. Ant.	On Dieulafoy	20 × 20	Ilc	sig	mp	Benign	HSE
8	Leone [4]	1995	41	M	C. Post.	On Dieulafoy	15	Ilc	sig	m	Benign	(-)
9	Yasutomo	1995	70	M	C. Ant.	On Dieulafoy	70 × 50	Ilc	tub 1	sm	Benign	Ethanol
10	Fuke	1996	34	F	C. Ant.	On Dieulafoy	25 × 25	Ilc + III	sig	sm	Ilc + III	Clipping + embo
11	Hisa	1997	71	M	C. Less.	On Dieulafoy	30 × 20	Ilc	tub 2	m	Benign	Clipping
12	Wakahara [2]	2000	56	M	M. Post.	On Dieulafoy	20 × 20	Ilc	tub 1	sm	Benign	Thrombin
13	Shimomatsuya	2000	42	F	C. Less.	On Dieulafoy	40 × 20	Ilc	tub 2	sm	Ilc	Clipping
14	Ikeda	2001	45	M	M. Ant.	On Dieulafoy	10	Ilc	sig	m	Ilc	Ethanol + HSE
15	Kishikawa [5]	2003	48	M	C. Post.	On Dieulafoy	Shallow	Ilc	sig	sm?	Benign	Heater-probe
16	Kishimoto [3]	2005	56	M	C. Less.	On Dieulafoy	50 × 25	Ilc + Ila	tub 2	mp	Benign	Clipping
17	Our case	2006	69	M	C. Post.	On Dieulafoy	50 × 40	Ila	tub 1	sm	Benign	Clipping

C, upper third; M, middle third; ant, anterior wall; less, lesser curvature; Ca, cancer; tub, tubular adenocarcinoma; tub 1, well-differentiated adenocarcinoma; tub 2, moderately differentiated adenocarcinoma; sig, signet-ring cell carcinoma; m, mucosa; sm, submucosa; mp, proper muscle; HSE, hypertonic saline epinephrine solution; endo. treatment, endoscopic treatment; embo, arterial embolization

ulcer nor deformity on the duodenal bulb, in spite of the patient's past history of duodenal ulcer.

The characteristic histopathological findings of Dieulafoy's artery were reported to be arteriovenous malformation, aneurysm, or a caliber-persistent artery [9–11]. We found an enlarged and tortuous artery in the submucosa which corresponded to a caliber-persistent artery. No abnormal vessels other than this caliber-persistent artery were found in the gastric body in our patient, but, unfortunately, serial sectioning was performed only around the cancer and in some other parts. Additionally, near the ulcer scar, the artery was obstructed with thickened intima caused by the endoscopic hemostasis. We provide a diagram of the abnormal vessels, looked at three-dimensionally, according to the serial sectioning of the resected specimen, in Fig. 3f.

Dieulafoy's ulcer coexisting with gastric cancer is rare. Since Sasaki [12] first reported this coexistence in 1982, 15 cases have been reported in Japanese journals [2,3], and 2 cases in English-language articles [4,5] (Table 1). Even though slightly depressed cancer; namely, type Ilc, was the most frequent type in the reported cases, we encountered a slightly elevated cancer, type Ila, and well-differentiated adenocarcinoma. It is difficult to answer the question why the Dieulafoy's ulcer coexisted with gastric cancer. However, gastric cancer was observed just above the Dieulafoy's lesion in all the reported patients. The following process is our speculation on the pathogenesis of this coexistence; however, we did not find any evidence other than in the reported cases. Circulation disturbance in Dieulafoy's vessels would produce repeated mucosal erosions and ulcers. Active regeneration and dysplasia of the mucosal membrane would be a factor promoting gastric cancer. This idea can explain why the cancer was located just above the Dieulafoy's ulcer.

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