A Caliber Persistent Artery (Dieulafoy’s Lesion) which is Associated with an Early-Stage Gastric Stump Cancer Following a Distal Gastrectomy

ABSTRACT
A 75–years old man was hospitalized with symptoms which suggested gastric cancer. Thirty-eight years ago, he had undergone a Billroth-II gastric reconstruction for a peptic ulcer. At the present admission, he had presented with an eight-month history of recurrent haematemesis, epigastric pain, vomiting, and fatigue. The emergent endoscopy showed a type 0-Iic (superficial depressed) early gastric stump cancer in the antrum, was admitted to our hospital with an eight-month history of recurrent haematemesis, vomiting and fatigue. On physical examination, normal bowel sounds and a soft and a nontender abdomen were described. No alcohol, tobacco, or other drug use was declared. No endoscopical surveillance was performed. His serology showed slight anaemia (haemoglobin 11 g/dl and haematocrit 40%), without any other blood disorders. The emergency upper gastrointestinal endoscopy which was performed, revealed a superficial depressed tumour in the antrum, which was located at 5.5 cm from the oesophagogastric junction. On cut section, corresponding to and surrounding the tumour, haemorrhagic areas were observed in the gastric wall.

Case Report

A 75–years old normostenic man (his body weight was 65 kg and his height was 1.63 m) who underwent distal gastrectomy with a Billroth-II reconstruction 38 years ago for a peptic ulcer which was located in the antrum, was admitted to our hospital with an 8–month history of recurrent haematemesis, vomiting and fatigue. In some cases, Dieulafoy’s lesion can masquerade other lesions such as a gastric cancer, that can produce severe and even fatal haemorrhages. It is either diagnosed at endoscopy or under a microscope, and its rarity is rather because of a lack of recognition than the true rarity. This lesion, which is also known as a gastric caliber-persistent artery, was first described in 1898 by Dieulafoy, as “exulceratio simplex” [1-3]. It is considered to be a vascular malformation of the left gastric artery, that consists of the persistence of the arterial diameter in the gastric wall, but the real pathogenesis is still under debate. Macroscopically, it is either an ulcerated or polypoid lesion which is usually localized within 6 cm of the proximal stomach, on the lesser curvature [1,4] but it has also been described in the oesophagus, duodenum, ileum, and the colon [3]. Under the microscope, a protruded or an ulcerated lesion with spurring arteries in its base is observed. Abnormally dilated vascular spaces are seen in the submucosa and the mucosa, and their erosion can produce severe bleedings [5,6]. On physical examination, normal bowel sounds and a soft and a nontender abdomen were described. No alcohol, tobacco, or other drug use was declared. No endoscopical surveillance was performed. His serology showed slight anaemia (haemoglobin 11 g/dl and haematocrit 40%), without any other blood disorders. The emergency upper gastrointestinal endoscopy which was performed, revealed a superficial depressed tumour in the antrum, which was located at 5.5 cm from the oesophagogastric junction. On cut section, corresponding to and surrounding the tumour, haemorrhagic areas were observed in the gastric wall.

INTRODUCTION
Dieulafoy’s lesion is a rare lesion of the gastrointestinal tract, that can produce severe and even fatal haemorrhages. It is either diagnosed at endoscopy or under a microscope, and its rarity is rather because of a lack of recognition than the true rarity. This lesion, which is also known as a gastric caliber-persistent artery, was first described in 1898 by Dieulafoy, as “exulceratio simplex” [1-3]. It is considered to be a vascular malformation of the left gastric artery, that consists of the persistence of the arterial diameter in the gastric wall, but the real pathogenesis is still under debate. Macroscopically, it is either an ulcerated or polypoid lesion which is usually localized within 6 cm of the proximal stomach, on the lesser curvature [1,4] but it has also been described in the oesophagus, duodenum, ileum, and the colon [3]. Under the microscope, a protruded or an ulcerated lesion with spurring arteries in its base is observed. Abnormally dilated vascular spaces are seen in the submucosa and the mucosa, and their erosion can produce severe bleedings [5,6].

In some cases, Dieulafoy’s lesion can masquerade other lesions such as a gastric cancer, that can facilitate the cancer diagnosis in the early stages. In this paper, we are presenting an early gastric stump cancer (GSC) which was diagnosed 38 years after a distal gastrectomy was performed for a gastric ulcer. The particularity of this case lies in its association with Dieulafoy’s lesion. To date, about 17 cases of gastric carcinomas (primary gastric cancers) which were associated with Dieulafoy’s lesions have been reported, the last one being reported in 2006 [5]. However, most of them were published in Japanese and only three were reported in the English literature [1,4,5]. This is the first case report that has revealed the association between a GSC and a Dieulafoy’s lesion. Based on the microscopic features which correlated with the real pathogenesis is still under debate. Macroscopically, it is either an ulcerated or polypoid lesion which is usually localized within 6 cm of the proximal stomach, on the lesser curvature [1,4] but it has also been described in the oesophagus, duodenum, ileum, and the colon [3]. Under the microscope, a protruded or an ulcerated lesion with spurring arteries in its base is observed. Abnormally dilated vascular spaces are seen in the submucosa and the mucosa, and their erosion can produce severe bleedings [5,6].

CASE PRESENTATION
A 75–years old normostenic man (his body weight was 65 kg and his height was 1.63 m) who underwent distal gastrectomy with a Billroth-II reconstruction 38 years ago for a peptic ulcer which was located in the antrum, was admitted to our hospital with an 8–month history of recurrent haematemesis, vomiting and fatigue. Based on the microscopic features which correlated with the
reCURRENT HAEMATEMESIS, THE FINAL DIAGNOSIS WAS AN EARLY GSC WHICH WAS ASSOCIATED WITH DIEULAFOY’S LESION. THE POSTOPERATORY EVOLUTION WAS FAVOURABLE; NO ADJUVANT THERAPY WAS USED. OUR PATIENT PRESENTED NO COMPLICATIONS SIX MONTHS AFTER THE SURGERY.

DISCUSSION

Although gastric cancer presents a tendency to decline over time, it remains the second leading cause of the cancer-related deaths in the world, especially in the European countries, where most of the cases are diagnosed in the advanced stages. In contrast, in the Asian countries, a significant number of cases are diagnosed in the early stages, but the risk of the associated GSC following a remote gastric surgery seems to increase [7,8]. About 1–3% of the patients who undergo gastrectomies will present with cancer in the gastric remnant [8]. Despite the well-known risk, most of the remnant cancers are still diagnosed in the advanced stages, most of the early GSCs being reported in Japan [1,5,9].

Most of the studies revealed that GSCs occur after gastric surgeries which are done for both benign and malignant lesions, usually in elderly males who are aged about 65–70 years (M:F = 3:1), the interval between the initial gastrectomy and the GSC occurrence being variable. In回顾性分析，早期胃癌的最终诊断与Dieulafoy的病变相关。手术后预后良好，未使用辅助治疗。患者术后6个月无并发症。

**DISCUSSION**

Although gastric cancer presents a tendency to decline over time, it remains the second leading cause of cancer-related deaths in the world, especially in European countries, where most of the cases are diagnosed in the advanced stages. In contrast, in Asian countries, a significant number of cases are diagnosed in the early stages, but the risk of an associated GSC following a remote gastric surgery seems to increase [7,8]. About 1–3% of the patients who undergo gastrectomies will present with cancer in the gastric remnant [8]. Despite the well-known risk, most of the remnant cancers are still diagnosed in the advanced stages, most of the early GSCs being reported in Japan [1,5,9].

Most of the studies revealed that GSCs occur after gastric surgeries which are done for both benign and malignant lesions, usually in elderly males who are aged about 65–70 years (M:F = 3:1), the interval between the initial gastrectomy and the GSC occurrence being variable. In most of the cases, the patients undergo gastrectomies for both benign and malignant lesions, usually in elderly males who are aged about 65–70 years (M:F = 3:1), the interval between the initial gastrectomy and the GSC occurrence being variable. In the studied cases, the patients underwent gastrectomies for both benign and malignant lesions, usually in elderly males who are aged about 65–70 years (M:F = 3:1), the interval between the initial gastrectomy and the GSC occurrence being variable. In the present case, the patient underwent a distal gastrectomy for a peptic ulcer 38 years ago, and developed recurrent hematemesis with anemia. The histopathological examination revealed an early cancer in the anastomotic area with a Dieulafoy’s ulcer and a small vessel protruding from the base and into the area of the gastric fundus, consistent with a well-differentiated adenocarcinoma.
being longer in the cases that undergo surgery for benign diseases as compared to that in the malignant ones (30–32 vs 10–18 years) [7,9,10] and also that in the Billroth–II reconstruction being longer as compared to that in the Billroth–I reconstruction (30–32 vs 12–20 years) [9,10]. There are also anatomical differences with respect to the occurrence of GSC, it being usually located near the suture line and the remnant gastric wall in case of the Billroth–I reconstruction, respectively at the anastomotic area after a Billroth–II procedure [9,10]. Knowing these particularities is essential for a proper follow-up and for an early detection of a GSC; an efficient surveillance program supposes the annual endoscopic examination of the gastric remnant for about 10–12 years following a primary surgery, almost every second year after this period [9]. In the present case, in line with the literature data, a GSC occurred in a 75-year-old man, 38 years following a distal gastrectomy which was done for a peptic ulcer, with a Billroth–II reconstruction, in the anastomotic area. The early detection was not done to the proper surveillance, but rather to the associated haematemesis, as a result of the Dieulafoy’s lesion. The longer interval for the GSC occurrence in case of the benign lesions as compared to the malignant ones, could lead to a supposition that the surgical resection could be the favouring factor of the cancer, but the pathogenesis of a GSC following gastric cancer is more complex and it depends on the properties of the remnant mucosa [10]. Moreover, the Billroth–II procedure which was used in the past for the correction of the benign gastric lesions seems to favour the streaming of the gastric stump anastomosis and, consequently, the continuous bloating of the anastomotic area with bile acids [10,11], as important risk factors for the cancer genesis in this area.

In this case, the recurrent haematemesis was the main clinical symptom of the GSC. Recurrent or severe gastrointestinal bleeding was also the main symptom of the preoperative diagnosis in the other gastric tumours that were associated with the Dieulafoy’s lesion, as we have shown in [Table/Fig-2]; three gastric carcinomas [1,4,5] and three stromal tumours [3,6] were reported to be associated with the Dieulafoy’s lesion, in the English literature [Table/Fig-2 and 3].

Based on the particularities of the present case and the literature data, we conclude that a meticulous endoscopical surveillance with multiple biopsies should be the golden standard from the 10 to the 15 years after the initial surgery in both benign and malignant primary gastric lesions. Independent of the medical history, the Dieulafoy’s lesion and gastric cancer should be suspected in the patients with recurrent haematemesis and/or anaemia and even in normostolic patients with a good performance status.

ACKNOWLEDGEMENT
This work was partially supported by the L’Oreal-Unesco National Fellowship for women in science. The English-language manuscript was polished by the SPI Global Professional Editing Service.

REFERENCES