Synchronous Gastrointestinal Stromal Tumor and Adenocarcinoma at the Gastroesophageal Junction

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The synchronous existence of two different tumors in the gastrointestinal tract is uncommon. We report the case of a 75-year-old man who had a concurrent gastrointestinal stromal tumor and adenocarcinoma at the gastroesophageal junction. The two tumors arose at the same site but had distinct morphologies. The etiology of synchronous tumors is still unclear and their coexistence causes problems for the surgeon, oncologist and pathologist in terms of their diagnosis, treatment, and follow-up. We report a rare case of synchronous tumors and a review of the literature.

Key Words: adenocarcinoma, gastroesophageal junction tumor, gastrointestinal stromal tumor
(*Kaohsiung J Med Sci* 2009;25:338–41)

The synchronous existence of two different tumors in the gastrointestinal tract is uncommon. Most cases involve adenocarcinomas, lymphomas, carcinoids or leiomyosarcomas of the stomach [1]. Concurrent gastrointestinal stromal tumors (GIST) and adenocarcinomas are extremely rare, with very few reported cases [2–5]. To the best of our knowledge, there has been no previous report of synchronous GIST at the gastroesophageal junction. Here we present an incidental finding of synchronous GIST and adenocarcinoma at the gastroesophageal junction.

Case Presentation

A 75-year-old man with a 1-month history of epigastralgia and anorexia was admitted to our hospital due to progressive symptoms. Physical examination revealed no obvious abnormalities and no lymphadenopathy. Laboratory data showed no anemia and the level of carcinoembryonic antigen was normal. However, esophagogastroduodenoscopy revealed a polyp-like mass about 1 cm in diameter over the gastroesophageal junction area (Figure 1). Computed tomography showed an eccentric tumor over the junction with no evidence of any other metastatic lesions.

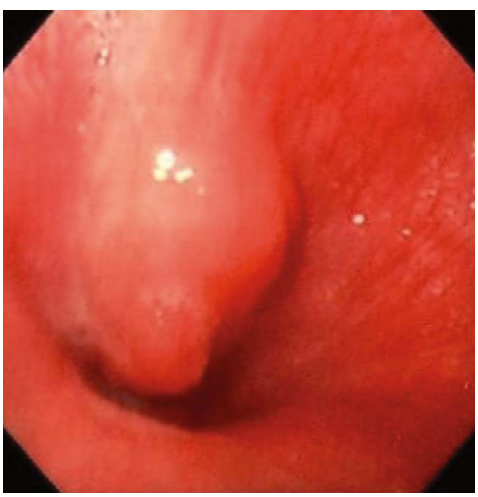

Figure 1. *Esophagogastroduodenoscopy revealed a polyp-like mass in the gastroesophageal junction area.*

Received: Nov 14, 2008 Accepted: Feb 25, 2009
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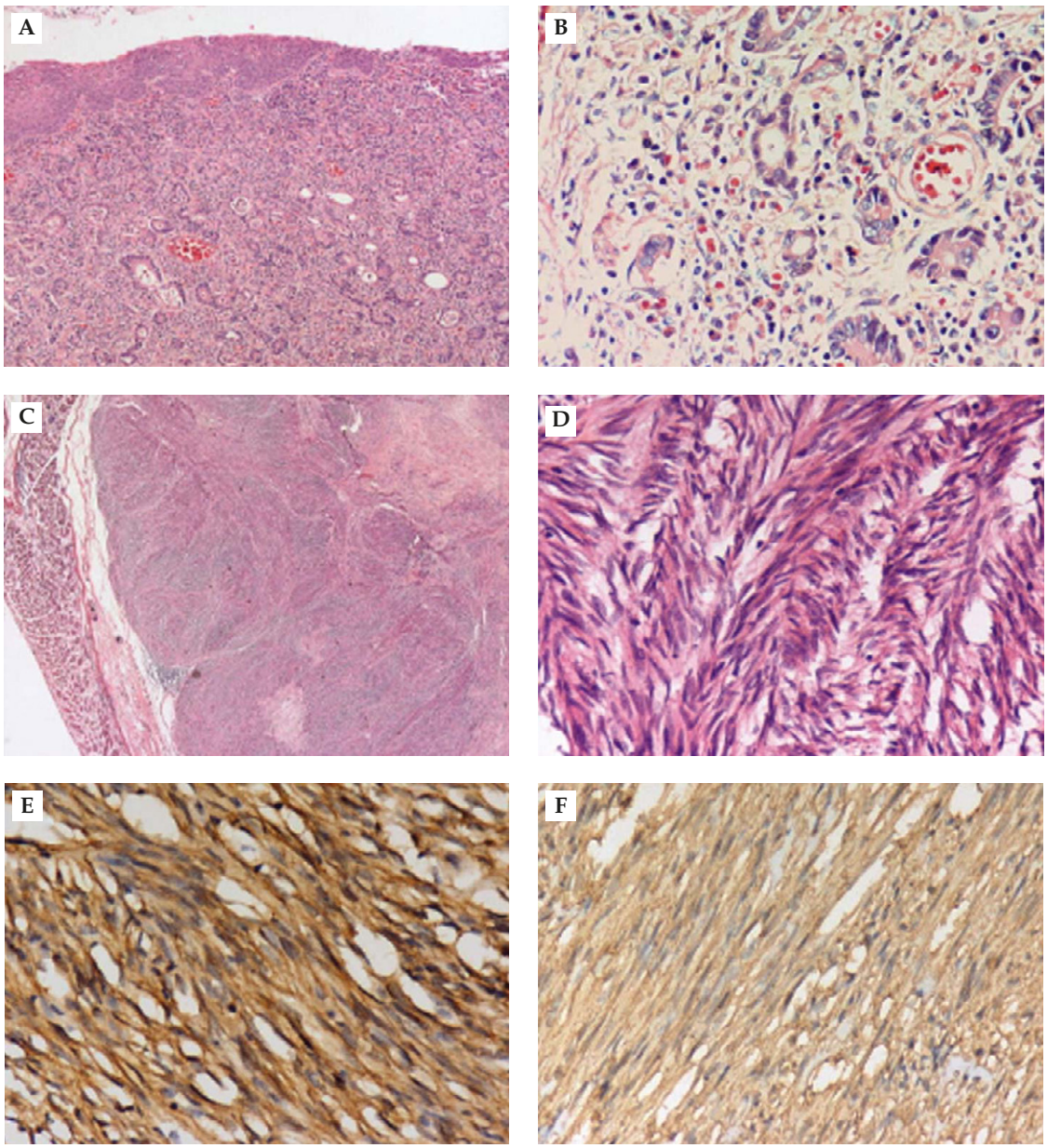

Figure 2. *Histologic findings. Gastroesophageal junction tumor infiltrated into submucosa: (A) low magnification; (B) high magnification. Several embedded nodules with spindle-shaped tumor cells were found in the gastric wall: (C) low magnification; (D) high magnification. The neoplastic cells were positive for: (E) CD117; (F) CD34.*

Biopsy showed a well-differentiated adenocarcinoma. The patient underwent proximal gastrectomy plus distal esophagectomy and pyloroplasty for suspected gastroesophageal junction tumor.

Gross examination of the gastroesophageal junction revealed an eroded lesion measuring 0.8 × 0.5 × 0.2 cm. In addition, there were five well-defined smooth nodular masses measuring up to 3.3 × 2.2 × 1.8 cm on the anterior and posterior serosal surfaces of the stomach. Serial sections showed the nodular masses to be located near the mucosa lesion but confined to the muscularis propria, without invasion of the overlying mucosa.

Histologic examination showed a well-differentiated adenocarcinoma infiltrating throughout the submucosa of the gastroesophageal junction (Figures 2A and 2B). No signs of lymphovascular permeation or perigastric lymph node metastasis were found. No signs of *Helicobacter pylori* infection were detected in the specimen. However, the embedded nodules in

the gastric wall, which were different from the adenocarcinoma at the gastroesophageal junction, were composed of distinct, spindle-shaped tumor cells (Figures 2C and 2D). The neoplastic cells were positive for CD117 and CD34 staining (Figures 2E and 2F), but negative for smooth muscle actin staining. Mitotic figures were rare within the tumors (average 2 mitoses/50 high-power fields). A diagnosis of synchronous GIST and adenocarcinoma at the gastroesophageal junction was made on the basis of these examinations.

Following total tumor resection, no recurrence of disease was detected by subsequent imaging studies and re-biopsy. Unfortunately, the patient suffered a brain infarction with hemorrhage 6 months later and died of respiratory failure due to pneumonia.

Discussion

The synchronous existence of two different tumors in the gastrointestinal tract is uncommon. Most cases involve adenocarcinomas, lymphomas, carcinoids, or leiomyosarcomas of the stomach [1]. Cases of synchronous GIST and adenocarcinoma, especially in the gastroesophageal junction area, have rarely been reported [2–5]. The current case also appears to be unique in that the larger GIST, rather than the small lesion of the adenocarcinoma, apparently accounted for the polyp-like lesion and symptoms in this patient. Although the real mechanism responsible for the coexistence of these tumors is not clear, special carcinogens, *H. pylori* infection, or genetic mutation have been considered to induce the transformation of different cell lines of the gastrointestinal tract [3,5,6]. However, most reported cases have involved older patients and in the high-risk regions for gastrointestinal tumors, which makes the coincident occurrence of both tumors less unlikely [2–5].

GISTs are rare, soft-tissue sarcomas arising primarily from mesenchymal tissues in the alimentary tract and the abdomen [6]. They are distinguished from other mesenchymal tumors by their unique expression of c-kit protein (CD117) [6,7]. Surgery has so far been the only effective treatment for GIST [6]. Recently, however, imatinib mesylate, a selective tyrosine kinase inhibitor, has shown some activity in GISTs expressing CD117 [8].

Although the coexistence of GIST with other tumors is rare and they tend to be small, asymptomatic, and occur in low-risk patients, it still causes problems for the surgeon, oncologist, and pathologist [9,10]. An accurate diagnosis is not possible if doctors do not examine samples thoroughly, especially in the case of small GISTs such as that in the current case. Second, surgical plans may need to be changed due to the incidental discovery of an unexpected tumor during surgery. Third, sufficient sampling is required to allow the diagnosis of suspicious metastatic/recurrent lesions during follow-up.

In summary, we report a rare case of synchronous GIST and adenocarcinoma at the gastroesophageal junction. Although the clinical course and behavior of this rare condition are still unclear, these cases highlight the importance of careful examination of samples and long-term follow-up.

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合併胃腸道基質瘤及腺癌的胃食道交接處腫瘤

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在胃腸道上合併兩種不同的腫瘤是少見的情形，我們報告一位七十五歲的男性病人，於胃食道交接處同時合併有胃腸道基質瘤及腺癌的病例。這兩種腫瘤是位於同一個位置，但有不同的獨特形態。同時合併腫瘤的形成機轉仍不明，但同時出現的腫瘤，不論是在診斷、治療及追蹤上，對於臨床的外科醫師、腫瘤科醫師及病理科醫師都是一種挑戰。我們報告此一病例並作文獻回顧。

關鍵詞：腺癌，胃食道交接處腫瘤，胃腸道基質瘤

(高雄醫誌 2009;25:338–41)

收文日期：**97** 年 **11** 月 **14** 日

接受刊載：**98** 年 **2** 月 **25** 日

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