Int J Clin Exp Pathol 2016;9(1):363-366
www.ijcep.com /ISSN:1936-2625/IJCEP0018196

Case Report

Gastrointestinal stromal tumor with synchronous gastric cancer: report of a case and review of literature

Gongting Zhou[1*], Neng Lou[2*], Ling Wu[3], Huajun Yu[1], Qiyu Zhang[1]

Departments of [1]Hepatobiliary Surgery, [2]Gastrointestinal Surgery, [3]Pathology, The First Affiliated Hospital of Wenzhou Medical University, Wenzhou, Zhejiang, P. R. China. [*]Equal contributors.

Received October 20, 2015; Accepted November 28, 2015; Epub January 1, 2016; Published January 15, 2016

Abstract: Gastrointestinal stromal tumors (GISTs) are mesenchymal tumors of the gastrointestinal tract with immunohistochemical reactivity for CD117 antibody. Gastrointestinal stromal tumor with synchronous gastric cancer is rare. A 69-year-old male presented with one-week history of epigastrium pain and vomit was admitted to our hospital. Subtotal gastrectomy and D2 lymph node dissection were performed, and reconstruction was provided with Billroth I procedure. Pathological examination of the specimens revealed gastrointestinal stromal tumor and poor-differentiated gastric cancer, respectively.

Keywords: Gastrointestinal stromal tumor, gastric cancer, stomach

Introduction

Gastrointestinal stromal tumors (GISTs) are malignant or potentially malignant tumors that arise from mesenchymal cells of the gastrointestinal tract [1]. The first case of synchronous epithelial and stromal tumors was described in 2000 [2]. Since then, only less than twenty cases have been described in case studies and less than 100 cases have been reported totally in the literature. However, its clinicopathological features are vague. Its preoperative diagnosis rate is low and the prognosis may be influenced by the correct diagnosis. Here we present a rare case of gastrointestinal stromal tumor with synchronous gastric cancer and provide a literature review.

Case presentation

A 69-year-old Chinese male with one-week history of upper abdominal pain and vomiting was admitted to our hospital in January 2013. He denied any history of abdominal discomfort. The physical examination and routine laboratory tests upon admission were unremarkable. However, an upper gastrointestinal endoscopy showed a mucosal ulceration located on the antrum (**Figure 1A**) and a polypoid mass located in the distal body with intact overlying mucosa (**Figure 1B**). High-grade intraepithelial neoplasia and early gastric cancer were verified with biopsies of mucosal ulceration. The polypoid mass was suspected of gastric polyp without biopsy. No metastatic lesions were found in either the abdominal ultrasonography or the CT scan. The patient underwent a distal subtotal gastrectomy, with D2 lymphadenectomy, and reconstruction was provided with Billroth I procedure. Histopathologic examination of the specimen of mucosal ulceration disclosed a poorly differentiated gastric cancer (**Figure 2A**) measuring 3×2.5 cm, infiltrating into the muscular layer. Interestingly, pathology of the polypoid mass measuring 1×1 cm, locating in the distal body, disclosed whirling sheets of spindle cells (**Figure 2B**). Immunohistochemical staining showed that the spindle cells were positive for CD117. No lymph node metastasis was detected in all retrieved lymph nodes. The staging of synchronous gastric cancer was Stage IB. The patient underwent regular clinical and radiological follow-up every 4 months after discharge. No evidence of tumor recurrence was found after 20 months of follow-up.

Discussion

The incidence of GIST is about 10 to 20 per 1,000,000 per year [3, 4]. The most common

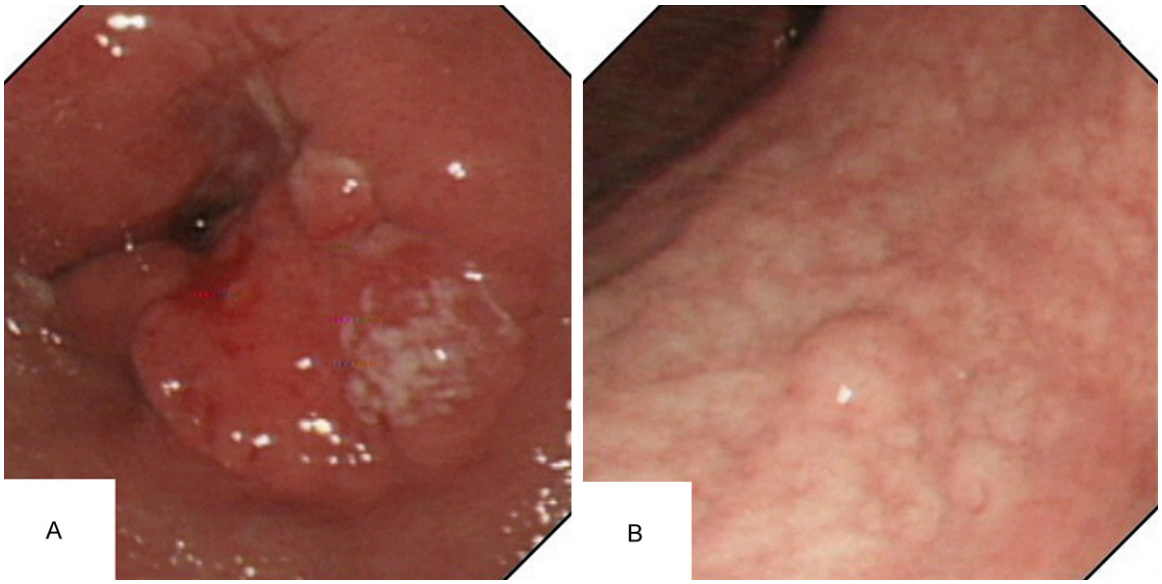

Figure 1. A: Endoscopic image of a mucosal ulceration with bleeding located on the antrum. B: Gastrointestinal endoscopy showed a polypoid mass located in the distal body with intact overlying mucosa.

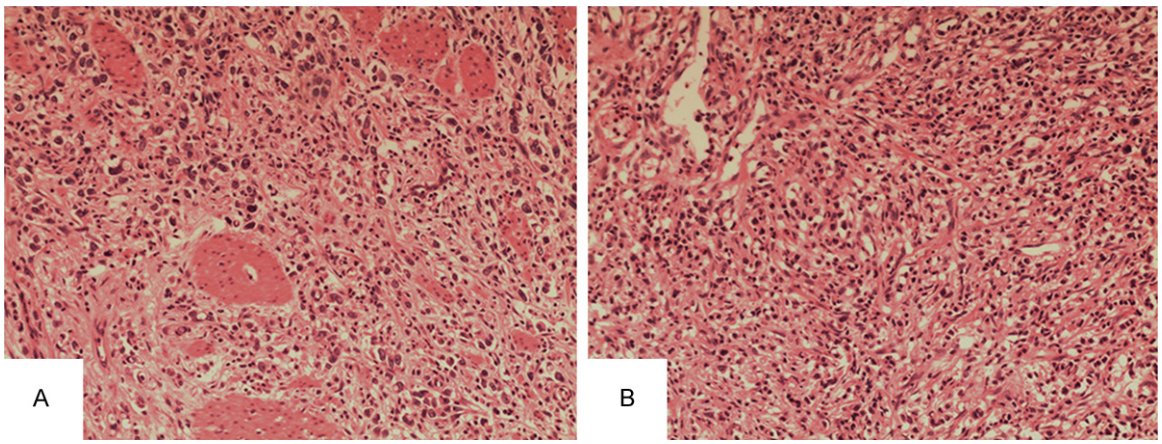

Figure 2. A: HE staining of the tumor showing poorly differentiated gastric adenocarcinoma cell (H&E, × 100). B: Photomicrograph from the resected polypoid mass showed whirling bundles of spindle cells with mitosis (H&E, × 100).

sites of GISTs are the stomach (60-84.8%), small intestine (10.5-30%), esophagus (1.2-5%), colon and rectum (3.5-5%) [1]. The coexistence of gastrointestinal stromal tumor with synchronous gastric cancer has been rarely reported. The preoperative diagnosis of GISTs depended mainly on imaging examination and biopsy [5-7]. However, gastric GISTs were often submucosal, muscular, or subserosal, with correctly preoperative diagnosis missed [8]. Clinical symptom of gastric GIST depends on tumor size and location. Many patients have no specific clinical symptoms when the tumor was small [1, 9]. Moreover, the clinical symptoms of gastric GIST with synchronous gastric cancer may be confused [1, 7, 9, 10].

In this case, the patient presented with one-week history of upper abdominal pain and vomiting. The symptoms attracted clinician's more attention on gastric cancer located on the antrum. Biopsy examination of the polypoid mass located in the distal body was missing. Not only the neglect of biopsy frequently occurs, but also the covering gastric mucosa may often remain intact and the endoscopic biopsies can

Table 1. Clinical features of 13 gastric GIST with synchronous gastric cancer patients

Author	Age	Sex	Synchronous tumors	Tumor position	Tumor size (cm)	Outcome
Lin [1]	70	F	RCC	Gastric angle	1.7 × 1.4	14 M Alive
de Roover [9]	70	M	GA	Antrum	3 × 3	24 M Alive
Villias [6]	78	M	GA	Anterior gastric body	NM	9 D Alive
Özgür [8]	63	M	GA	Cardia	9	13 M Died
Özgür [8]	60	M	GA	Antrum	4	12 M Alive
Theodosopoulos [15]	80	M	GA	Antrum	6 × 4	12 M Alive
Kleist [7]	78	M	GA	Lessor curvature	6 × 5.5	4 M Died
Kleist [7]	86	F	GA	Minor curvature	6	11 M Alive
Bi [16]	73	F	GA	Fundus and body	4 × 3	NM
Antonio [17]	68	M	GA	Lessor curvature	6.5 × 3.8	6 M Alive
Sailors [18]	65	F	GA	Lessor curvature	1 × 1.5	NM
Liu [19]	70	M	GA	Cardia and body	8 × 5 × 3	3 M Died
Lee [11]	82	M	RCC	Gastric body	8 × 6 × 6	NM

GIST, gastrointestinal stromal tumor; GA, gastric adenocarcinoma; RCC, ring cell carcinoma; D, days; M, months; NM, not mentioned.

be inefficient [11, 12]. Subsequently, the correct diagnosis was made from postoperative histopathologic examination and immunohistochemical staining.

The correctly preoperative diagnosis of such cases can be a challenge. Lin *et al.* [12] retrospectively investigated clinical features of 42 patients of gastric gastrointestinal stromal tumor with synchronous gastric cancer and his study showed that 76.2% of patients are male and 71.4% of patients are over 60 years old. Surprisingly, only one tumor (2.4%) was detected before surgery. In present study, after performing a literature search, we also reviewed the clinical data of 13 patients with gastrointestinal stromal tumor and synchronous gastric cancer, and the details are showed in **Table 1**. The mean age was 72.3 years old and ranged from 60 to 86. Similarly, the patients with gastrointestinal stromal tumor and synchronous gastric cancer were seem old in age. As the pace of global ageing is impressive, such cases would not be rare. The histological subtype of the synchronous gastric cancer included adenocarcinoma and ring cell carcinoma of which incidence needs larger sample research to clarify. The rate of correct preoperative diagnosis of the 13 cases is low, and most of them were diagnosed postoperatively.

The study in Japan showed that the three years overall survival (OS) rate was 62.6%, in a 42 patients group with gastric GIST and synchronous gastric cancer [12]. Although the rate of correct preoperative diagnosis of GIST in such combination is low, the prognosis is not so disappointing. Interestingly, gastric cancer plays a major role on survival in such combination [12]. Kazuyoshi *et al.* [13] reported that the Japanese gastric cancer screening system could help early detect gastric GIST. Besides, enhancing awareness of clinician about multiple primary tumors may also raise the rate of correct diagnosis.

In spite of complete resection of the GISTs, GISTs may relapse and end in poor prognosis [14]. Adjuvant chemotherapy and targeted therapy are important for high-risk GISTs [12]. Therefore, awareness of such combination is important to make accurate and effective treatment strategy. More cases are required for evaluating the relationship and the tumorigenesis of such combination.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Qiyu Zhang, Department of Hepatobiliary Surgery, The First Affiliated Hospital of Wenzhou Medical University, Fu-Xue Xiang, Wenzhou 325000, Zhejiang, P. R. China. E-mail: zqydoc@163.com

References

[1] Lin YL, Tzeng JE, Wei CK and Lin CW. Small gastrointestinal stromal tumor concomitant

with early gastric cancer: a case report. World J Gastroenterol 2006; 12: 815-817.
[2] Maiorana A, Fante R, Maria Cesinaro A and Adriana Fano R. Synchronous occurrence of epithelial and stromal tumors in the stomach: a report of 6 cases. Arch Pathol Lab Med 2000; 124: 682-686.
[3] Nilsson B, Bumming P, Meis-Kindblom JM, Oden A, Dortok A, Gustavsson B, Sablinska K and Kindblom LG. Gastrointestinal stromal tumors: the incidence, prevalence, clinical course, and prognostication in the preimatinib mesylate era--a population-based study in western Sweden. Cancer 2005; 103: 821-829.
[4] Tzen CY, Wang JH, Huang YJ, Wang MN, Lin PC, Lai GL, Wu CY and Tzen CY. Incidence of gastrointestinal stromal tumor: a retrospective study based on immunohistochemical and mutational analyses. Dig Dis Sci 2007; 52: 792-797.
[5] Paiva CE, Moraes Neto FA, Agaimy A, Custodio Domingues MA and Rogatto SR. Perivascular epithelioid cell tumor of the liver coexisting with a gastrointestinal stromal tumor. World J Gastroenterol 2008; 14: 800-802.
[6] Villias C, Gourgiotis S, Veloudis G, Sampaziotis D and Moreas H. Synchronous early gastric cancer and gastrointestinal stromal tumor in the stomach of a patient with idiopathic thrombocytopenic purpura. J Dig Dis 2008; 9: 104-107.
[7] Kleist B, Lasota J and Miettinen M. Gastrointestinal stromal tumor and gastric adenocarcinoma collision tumors. Hum Pathol 2010; 41: 1034-1039.
[8] Firat O, Caliskan C, Karaca C, Sezak M, Ozutemiz O, Ersin S and Guler A. Coexistence of gastric cancer and gastrointestinal stromal tumor: report of two cases. Turk J Gastroenterol 2010; 21: 302-304.
[9] de Roover A, Detry O, de Leval L, Coimbra C, Desaive C, Honore P and Meurisse M. Report of two cases of gastric cancer after bariatric surgery: lymphoma of the bypassed stomach after Roux-en-Y gastric bypass and gastrointestinal stromal tumor (GIST) after vertical banded gastroplasty. Obes Surg 2006; 16: 928-931.
[10] Kim SH, Lee ST, Jeon BJ, Kim IH, Kim SW, Lee SO, Kim DG and Park HS. Signet-ring cell carcinoma mimicking gastric gastrointestinal stromal tumor confirmed by endoscopic ultrasound-guided trucut biopsy. Clin Endosc 2012; 45: 421-424.
[11] Lee FY, Jan YJ, Wang J, Yu CC and Wu CC. Synchronous gastric gastrointestinal stromal tumor and signet-ring cell adenocarcinoma: a case report. Int J Surg Pathol 2007; 15: 397-400.
[12] Lin M, Lin JX, Huang CM, Zheng CH, Li P, Xie JW, Wang JB and Lu J. Prognostic analysis of gastric gastrointestinal stromal tumor with synchronous gastric cancer. World J Surg Oncol 2014; 12: 25.
[13] Yamamoto K, Tsujinaka T, Takahashi T, Sato S, Nishiguchi Y, Nakashima Y, Muguruma K, Hirota S and Nishida T. Impact of the Japanese gastric cancer screening system on treatment outcomes in gastric gastrointestinal stromal tumor (GIST): an analysis based on the GIST registry. Ann Surg Oncol 2015; 22: 232-239.
[14] Hassan I, You YN, Shyyan R, Dozois EJ, Smyrk TC, Okuno SH, Schleck CD, Hodge DO and Donohue JH. Surgically managed gastrointestinal stromal tumors: a comparative and prognostic analysis. Ann Surg Oncol 2008; 15: 52-59.
[15] Theodosopoulos T, Dellaportas D, Psychogiou V, Gennatas K, Kondi-Pafiti A, Gkiokas G, Papaconstantinou I and Polymeneas G. Synchronous gastric adenocarcinoma and gastrointestinal stromal tumor (GIST) of the stomach: a case report. World J Surg Oncol 2011; 9: 60.
[16] Bi R, Sheng W and Wang J. Collision tumor of the stomach: gastric adenocarcinoma intermixed with gastrointestinal stromal tumor. Pathol Int 2009; 59: 880-883.
[17] Antonio L, Guzman P, Villaseca M, Araya J, De Toro G and Roa J. [Concomitant presence of a gastric adenocarcinoma and gastrointestinal stromal tumor: report of one case]. Rev Med Chil 2009; 137: 531-536.
[18] Sailors JL and French SW. The unique simultaneous occurrence of granular cell tumor, gastrointestinal stromal tumor, and gastric adenocarcinoma. Arch Pathol Lab Med 2005; 129: e121-123.
[19] Liu SW, Chen GH and Hsieh PP. Collision tumor of the stomach: a case report of mixed gastrointestinal stromal tumor and adenocarcinoma. J Clin Gastroenterol 2002; 35: 332-334.