Gastrointestinal stromal tumor of the stomach with a giant abscess penetrating the gastric lumen

Taro Osada, Akihito Nagahara, Tomohiro Kodani, Akihiro Namihisa, Masato Kawabe, Takashi Yoshizawa, Toshifumi Ohkusa, Sumio Watanabe

Abstract
Gastrointestinal stromal tumors (GISTs) are the most common mesenchymal tumors of the gastrointestinal tract. In large GISTs, cystic degeneration, necrosis and focal hemorrhage that occur inside the tumor can result in gastrointestinal bleeding. We describe a case of a 74-year-old male with GIST of the stomach accompanied with a giant abscess that penetrated the gastric lumen. The patient experienced undiagnosed fever for two months prior to hospitalization. Gastrointestinal endoscopy, X-ray series and computed tomography of the patient’s abdomen revealed a gastric submucosal tumor in the fornix, with a fistula to the gastric lumen that was inundated with a great deal of pus. The mass was diagnosed as a GIST from biopsy specimens. The patient was treated by endoscopic drainage of the abscess and intravenous administration of antibiotics. Eventually, a partial gastrectomy was performed. He was also administered Imatinib mesylate as adjuvant therapy. He was followed up for 2 years and no metastasis or recurrence was recognized at the follow-up examinations. This is the first report of a patient with GIST of the stomach and a giant abscess who experienced undiagnosed fever for two months prior to hospitalization.

© 2007 The WJG Press. All rights reserved.

Key words: Gastrointestinal stromal tumor; Abscess; Drainage

INTRODUCTION
Gastrointestinal stromal tumors (GISTs), which arise primarily in the gut wall, are mesenchymal neoplasms closely related to the interstitial cells of Cajal. GISTs typically carry gain-of-function mutations in genes encoding the KIT receptor tyrosine kinase (CD117) or platelet-derived growth factor receptor α, both of which are involved in cell survival, development and proliferation. Although GISTs can arise at any location in the gastrointestinal tract, they are found most often in the stomach (60%-70%), followed by the small intestine (20%-30%), colon and rectum (5%), and esophagus (< 5%).

Bacteria-induced abscess in the gastric wall is extremely rare, and the diagnosis is often late. Prior to the development of sufficient therapy, suppurative gastritis often results in death, with a mortality rate reported between 37% and 84%. Here, we report an unusual case of a patient with GIST of the stomach and a giant abscess who experienced undiagnosed fever for two months prior to hospitalization.

CASE REPORT
A 74-year-old man presented with fever and abdominal pain in the past two months. On examination, there was fever (38.1°C) and minimal tenderness in the epigastrium, but no jaundice or abnormal lymph node enlargement. The patient was taking no medications and his past medical history was unremarkable. Routine laboratory data on admission showed a white blood cell count of 10,000/mm³, C-reactive protein of 16.5 mg/dL and an erythrocyte sedimentation rate of 65 mm/h. Other chemistry and liver function tests were normal.

Gastrointestinal endoscopy revealed a gastric submucosal tumor in the fornix, with a fistula to the gastric lumen that was inundated with a great deal of turbid, yellow fluid (Figure 1). The fluid was found to contain abundant Streptococcus intermedius. Examination of the upper gastrointestinal tract X-ray series that included administration of amidotrizoic acid (Gastrografin® Nihon Schering K.K. Osaka Japan) revealed a large cavity, about...
7 cm × 8 cm, penetrating into the gastric lumen (Figure 2). An enhanced computed tomography (CT) scan of the abdomen revealed a large central cavity in the tumor that was partially filled with fluid, and irregular thickening of the gastric wall in the posterior aspect of the fundus (Figure 3). No other visceral abnormalities were found.

Biopsy specimens taken from the fistula edge during endoscopy resembled a gastric myogenic tumor composed of spindle cells. Immunohistochemical staining for KIT and CD34 was positive, whereas staining for α-smooth-muscle actin, desmin and S-100 protein was negative. The labeling index (L.I) for MIB-1, determined by counting positively stained nuclei among 1000 tumor cells, was about 18% (Figure 4).

The mass was diagnosed as a GIST accompanied with a giant abscess. The patient was treated by endoscopic drainage of the abscess through the fistula and intravenous administration of antibiotics. Subsequent to improvement in his clinical condition and laboratory tests, a partial gastrectomy (proximal partial gastrectomy) was performed 20 d after admission. Macroscopic examination revealed a 10 cm × 12 cm × 7 cm submucosal, intramural tumor protruding from the serosal surface (Figure 5). The cut surfaces were grayish-white and showed a large amount of central necrosis with a fistula to the gastric lumen. The surgical margins were free of tumor.

DISCUSSION

GISTs are the most common mesenchymal tumors of the gastrointestinal tract[8]. In general, tumor size and mitotic index are accepted as two independent prognostic factors for diagnosis[9]. For GISTs, prognostic markers that include size larger than 5 cm, mitotic rate > 5/50, high-power fields (HPF), tumor necrosis and a Ki-67 (MIB-1) index ≥ 10% all are associated with malignancy and high mortality[10,11]. For patients with large or mitotically active gastric GISTs, a MIB-1 proliferative index ≥ 10% is predictive of early recurrence and tumor-related death[12].

Whereas small GISTs (5 cm or less) can be treated by wedge gastric resection, larger tumors may require subtotal or total gastrectomy, including omentectomy. Metastatic or unresectable GISTs are treated with Imatinib mesylate (STI571), a small molecular tyrosine kinase inhibitor of KIT, platelet-delivered growth factor receptor and BCR-ABL. The large size (> 5 cm) and high mitotic count of the tumor in the present case classified it as a high risk. Since cystic degeneration, necrosis and focal hemorrhage generally predominate in larger tumor masses, we hypothesize that the central cavity in this GIST could be due to extensive necrosis.
Surpurative gastritis has generally been regarded as a secondary phenomenon caused by the hematogenous spread of bacteria from other infected organs or from a local invasion of the gastric mucosa\(^a\). Alcoholism, diabetes mellitus, decreased gastric acidity and immuno-suppression have been regarded as predisposing factors\(^b\). Suppurative gastritis can be classified as diffuse or localized. A gastric wall abscess is localized and less common, and frequently involves the antrum or pylorus\(^c\). In order of frequency, Streptococci, Staphylococci, E. coli, Haemophilus influenzae, Proteus and Clostridium are the most common pathogens\(^d\).

This particular case of gastric abscess also contained Streptococcus intermedius. A combination therapy that includes systemic administration of antibiotics and endoscopic drainage of pus has been reported to be an alternative to aggressive surgery\(^e\). Since no organ besides the stomach presented any sign of infection, the abscess in this case might have formed due to bacteria that spread to the adjacent cavity via the fistula. Endoscopic drainage of the abscess and administration of antibiotics resulted in the clinical improvement of this patient. He also was treated with Imanitib mesylate as adjuvant therapy, and no metastasis or recurrence was recognized during the 2-year follow-up period.

Few cases of submucosal tumor accompanied with abscess formation have been reported\(^11\). Recently, following endoscopic biopsy, a giant GIST with abscess formation was reported\(^12\). This GIST was large (12 cm × 13 cm in diameter) and accompanied with a closed abscess that did not penetrate into the gastric lumen. In 1998, Seidel et al\(^13\) reported a gastric leiomyosarcoma with abscess penetrating into the gastric lumen. This case of submucosal tumor is more similar to our case because spontaneous drainage of pus from the gastric abscess to the lumen was observed endoscopically. Because no immunohistochemical staining for KIT and CD34 was carried out, the case could not clearly be diagnosed as GIST. Hence, this is the first report of a patient with clearly diagnosed GIST accompanied with endoscopic evidence of an abscess penetrating into the gastric lumen.

REFERENCES

1. Hirota S, Itozaki K, Moriyama Y, Hashimoto K, Nishida T, Ishiguro S, Kawano K, Hanada M, Kurata A, Takeda M, Muhammad Tunio G, Matsuzawa Y, Kanakura Y, Shinomura Y, Kitamura Y. Gain-of-function mutations of c-kit in human gastrointestinal stromal tumors. Science 1998; 279: 577-580
2. Judson I. Gastrointestinal stromal tumors (GIST): biology and treatment. Ann Oncol 2002; 13 Suppl 4: 287-289
3. Miettinen M, Majdi M, Lasota J. Pathology and diagnostic criteria of gastrointestinal stromal tumors (GISTs): a review. Eur J Cancer 2002; 38 Suppl 5: S39-S51
4. Pidhorecky I, Cheney RT, Kraybill WG, Gibbs JF. Gastrointestinal stromal tumors: current diagnosis, biologic behavior, and management. Ann Surg Oncol 2000; 7: 705-712
5. Miller AL, Smith B, Rogers AL. Phlegmonous gastritis. Gastroenterology 1975; 68: 231-238
6. Starr A, Wilson JM. Phlegmonous gastritis. Ann Surg 1957; 145: 88-93
7. Stephenson SE, Yasrebi H, Rhatigan R, Woodward ER. Acute phlegmasia of the stomach. Am Surg 1970; 36: 225-231
8. Shimomura Y, Kinoshita K, Tsutsui S, Hirota S. Pathophysiologic, diagnosis, and treatment of gastrointestinal stromal tumors. J Gastroenterol 2005; 40: 775-780
9. Miettinen M, Monihan JM, Sarlomo-Rikala M, Kovatch AJ, Carr NJ, Emory TS, Sobin LH. Gastrointestinal stromal tumors/smooth muscle tumors (GISTs) primary in the omentum and mesentery: clinicopathologic and immunohistochemical study of 26 cases. Ann J Surg Pathol 1999; 23: 1109-1118
10. Emory TS, Sobin LH, Lukes L, Lee DH, O’Leary TJ. Prognosis of gastrointestinal smooth-muscle (stromal) tumors: dependence on anatomic site. Ann J Surg Pathol 1999; 23: 82-87
11. Hasegawa T, Matsuno Y, Shimoda T, Hirohashi S. Gastrointestinal stromal tumor: consistent CD117 immunostaining for diagnosis, and prognostic classification based on tumor size and MIB-1 grade. Hum Pathol 2002; 33: 669-676
12. Toquet C, Le Néel JC, Guillou L, Renaudin K, Hamy A, Heymann MF, Simon-Valla S, Le Borgne J, Maugard C, Fiche M. Elevated (KMPgt; or = 10%) MIB-1 proliferative index correlates with poor outcome in gastric stromal tumor patients: a study of 35 cases. Dig Dis Sci 2002; 47: 2247-2253
13. Murphy JF, Graham DY, Frankel NB, Spjut HJ. Intramural gastric abscesses. Am J Surg 1976; 131: 618-621
14. Berk RN, Reit RJ. Intra-abdominal chicken-bone abscesses. Radiology 1971; 101: 311-313
15. Vandyk K, Geman JD. Empyema of the gallbladder causing gastrointesdinal abscess and pyloric obstruction. Am J Surg 1967; 113: 295-297
16. Avilés JF, Fernández-Seara J, Bárdena R, Domínguez F, Fernández C, Ledo L. Localized phlegmonous gastritis: endoscopic view. Endoscopy 1988; 20: 38-39
17. Zazzo JF, Tócheg G, Millat B, Aubert A, Bedossa P, Kéros L. Phlegmonous gastritis associated with HIV-1 seroconversion. Endoscopic and microscopic evolution. Dig Dis Sci 1992; 37: 1454-1459
18. Will U, Masri R, Bossecert H, Knopke A, Schönlebe J, Justus J. Gastric wall abscess, a rare endosonographic differential diagnosis of intramural tumors: successful endoscopic treatment. Endoscopy 1998; 30: 432-435
19. Lantz PE, Westerman EL, Seifert RW. Gastric wall abscess drained at endoscopy. Gastrointest Endosc 1989; 35: 272-274
20. Nozawa S, Bando T, Nagata T, Tsukada K. Abscess formation in a giant gastrointestinal stromal tumor of the stomach following endoscopic biopsy. Endoscopy 2006; 38: 955
21. Seidel RH, Burdick JS. Gastric leiomyosarcoma presenting as a gastric wall abscess. Am J Gastroenterol 1998; 93: 2241-2244
22. Honda K, Mikami T, Ohkusa T, Takashimizu I, Fujiki K, Araki A, Shimoi K, Enomoto Y, Ariake K, Miyasaka N, Nihei Z, Oda K, Terada T. Gastrointestinal autonomic nerve tumor with giant abscess. A case report and literature review. J Clin Gastroenterol 1997; 24: 280-285
23. S-Editor Wang J, L-Editor Wang XL, E-Editor Che YB