Gastric Inflammatory Myofibroblastic Tumor with Fludeoxyglucose ($^{18}$F, FDG) Uptake on Positron Emission Tomography (PET): A Case Report and Literature Review

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Abstract

Background: Inflammatory Myofibroblastic Tumor (IMT) is a very rare neoplasm. When Endoscopic Gastroduodenoscopy (EGD) was performed, submucosal tumors of stomach were found incidentally. According to different sources of layer and echogenicity in the Endoscopic Ultrasonography (EUS) finding, submucosal tumors like carcinoid, pancreatic rest, Gastrointestinal Stromal Tumor (GIST), leiomyoma and schwannoma wound be distinguished. However, IMTs are one of submucosal tumors in the stomach. We must put this impression in the differential diagnosis when performing EUS.

Case Summary: We present a 55-year-old woman without symptoms who received a health examination, and had a gastric tumor was found during EGD. Initial biopsies showed chronic inflammation. Positron Emission Tomography (PET) showed an increased Fludeoxy Glucose (FDG) uptake in the stomach. Endoscopic ultrasonography was also performed. After surgical intervention, pathological analysis identified an inflammatory myofibroblastic tumor. No recurrence was observed by EGD or a PET scan during the follow-up. The relevant literature from the PubMed database was reviewed, and the clinical presentation, laboratory data, treatment strategies and outcomes of 42 reported cases were analyzed. Forty-two patients with gastric IMTs showed a female predominance (female: male: 26: 16). The most common location of gastric IMTs were gastric bodies (18 of 42). The most common symptoms were abdominal pain (21 of 42). Only two cases were asymptomatic. Tumor recurrence was found in 3 cases after surgical intervention in the reviewed literature.

Conclusion: EUS is useful to identify submucosal tumor in the gastrointestinal tract. IMTs must keep in mind when performing EUS.

Abbreviations: CHD: Coronary Heart Disease; ACS: Acute Coronary Syndrome; SCD: Stable Coronary Heart Disease; HbA1c: Hemoglobin A1c; Lp(a): Lipoprotein a; Fbg: Fibrinogen; Hcy: Homocysteine; CRP: C-Reactive Protein; AS: Atherosclerosis; TG: Triglyceride; FBG: Fasting Blood Glucose

Introduction

Inflammatory Myofibroblastic Tumor (IMT) is a very rare neoplasm usually seen in young people and children [1]. While the lung is the most common site [2], IMT is rarely observed in the stomach especially in adults. Gastric IMTs, which shows a female predominance, may present with a variety of symptoms depending on the location of the tumor. The difficulty in radiologically differentiating between malignant and benign lesions imposes another clinical challenge to clinicians. In this report, we present a case of a gastric IMT in an asymptomatic woman.
Case Presentation

Brief history

A 55-year-old woman without known systemic disease underwent a physical check-up in May 2014. Basic laboratory studies gave normal values (Hb: 12.3gm/dL, WBC: 7100/μL, INR: 1.09). Besides, liver and renal functions as well as levels of tumor markers including CEA, CA199, alpha-fetoprotein were within normal limits. There were no abdominal discomfort, no nausea or vomiting, no weight loss or other discomfort recently. No hypertension or diabetes mellitus were found in her family history. She was allergy to pyrine. No surgical history was found. On the other hand, PET scan showed increased Fludeoxy Glucose (FDG) uptake over the greater curvature of the stomach (Figure 1). Endoscopic gastroduodenoscopy revealed a reddish tumor with central depression about 1cm over the greater curvature of mid-body (Figures 2a & 2b). Endoscopic ultrasound study demonstrated focal thickening of muscular layer with loss of layering over the first, second and third layers (Figure 3) for which biopsy was taken. Pathologic analysis showed chronic inflammation with infiltration of lympho-plasma cells. The initial impression was Gastrointestinal Stromal Tumor (GIST) or leiomyoma. After discussion with the pathologist, IMT was diagnosed. We discussed with patient about treatment for this tumor. The patient agreed to receive therapeutic endoscopy for tumor resection. After a futile attempt of local endoscopic resection due to technical problem, surgical intervention of subtotal gastrectomy with gastroduodenostomy was performed (Figure 4). After stabilization of general condition, the patient was discharged with regular follow-ups at the outpatient clinic. There was no evidence of tumor recurrence after following the patient for 24 months by endoscopic gastroduodenoscopy and PET scan. No adverse and unanticipated events were found during follow-up.
Figure 2: Tumor features by endoscopic gastroduodenoscopy. A: A 1 cm mass was found above the greater curvature of the mid-body (white light view). B: It is suspected to be a submucosal tumor in the Narrow Band Imaging (NBI) view.

Figure 3: Submucosal tumor, up to 1cm in size, with focal thickening of the muscular layer and loss of layering over the 1st, 2nd and 3rd layers observed by endoscopic ultrasound (EUS).
Pathology

Microscopically, the tumor was mainly located at the submucosa with an infiltrating border. The overlying mucosa was ulcerated. The tumor was composed of bland-looking myofibroblasts and lympho-plasma cells within a fibrotic and collagenized stroma (Figure 5a). No abnormal mitotic figure or tumor necrosis was found. Immunohistochemical staining showed CD68-positive but ALK-negative myofibroblasts (Figure 5b). The diagnosis of an ALK-negative inflammatory myofibroblast tumor was made.

Figure 4: Frontal view of the tumor (white arrows).

Figure 5: A: Features of the IMT by microscopy. A: The tumor is composed of bland looking myofibroblasts and lympho-plasma cells within a fibrotic and collagenized stroma. B: ALK staining was negative.

Final Diagnosis

The final diagnosis was IMT which was origin from Muscularis propria layer.

Outcome and Follow-up

After stabilization to general conditions, the patient was discharged with regular follow-ups at the outpatient clinic. There
was no evidence of tumor recurrence after following the patient for 24 months by endoscopic gastroduodenoscopy and PET scan. No adverse and unanticipated events were found during follow-up.

**Discussion**

**Background Review**

Inflammatory Myofibroblastic Tumor (IMT), also known as inflammatory pseudotumor, is rare [3]. IMT is usually seen in children and young adults with a slight male predominance (male: female, 1:4) [4,5]. Gastric IMT, which has been found to show a female predominance (male: female = 1:4) [6,7], is even rarer. Although the lung is most commonly affected, IMT has been reported to involve other organs such as the liver, pancreas, spleen, lymph node, breast, kidney, bladder, orbits, and central nervous system [8]. It has been hypothesized that IMT is associated with uncontrolled inflammation from Epstein-Barr virus, human herpesvirus-8, E. coli, H. pylori, or cytomegalovirus infections as well as gastroesophageal reflux disease [9]. Moreover, recent studies imply that some IMTs are related to IgG4 [10]. Although IMT may be asymptomatic, symptoms related to its location might be observed [5]. IMT has been reported to be associated with fever, malaise, weight loss, anemia, and thrombocytosis but can also be asymptomatic [8], depending on the location of the tumor in the stomach [11]. Gastric IMT may also spread to adjacent organs and cause various symptoms, which usually subside after tumor resection.

**Literature Review**

All of the English literature in the PubMed database from January 1989 to December 2017 was searched using the key words “Positron emission tomography”, “Gastric inflammatory myofibroblastic tumor”, and “Inflammatory pseudotumor”. The demographic (i.e., age and gender) and clinical manifestations, laboratory findings, locations and sizes of tumor; pathological features of biopsied specimens, treatment strategies and follow-ups of the reported patients were reviewed. Non-English literature and reports on patients younger than 18 years of age were excluded. All statistical analyses were performed using commercially available SPSS software (version 15.0 for Windows; SPSS, Chicago, IL, Unites States). Data are expressed as the mean ± SD. The results are summarized in Table 1.

**Patient Demography and Clinical Characteristics of Gastric Imts**

A review of the patient demography, clinical manifestations, tumor characteristics, and treatments of 42 previously reported cases of gastric IMTs (Table 1) showed a female predominance (female: male, 26:16) and a mean age of 29.83±21.61 years (ranging from 4 months – 80 years old). The most common symptoms were abdominal pain, followed by vomiting (or nausea), body weight loss, hematemesis (or melena), fever, dysphagia, dyspepsia (or pyrosis), palpable abdominal mass, poor appetite or anorexia, and fatigue. The symptoms are related to the locations of the tumors. Laboratory studies revealed that anemia was the most common anomaly in these patients (22 out of 33). Other abnormalities included elevated Erythrocyte Sedimentation Rate (ESR), leukocytosis, elevated IgA, impaired liver function, elevated LDH, and elevated neuron-specific enolase. The most common location of tumors during endoscopic examinations was the gastric body (42.9%) (18 of 42). Other reported locations of gastric IMTs included the antrum, cardia, gastroesophageal junction, angle, and site of gastrojejunostomy. Chronic inflammation or inflammatory cells (5 of 13) was the most common finding upon pathological examination of biopsied specimens among the reviewed literature in which a biopsy was performed. (Table 2), possibly due to the subepithelial location of the tumor.

**Table 1:** Summary of the demographics and clinical characteristics of reported inflammatory myofibroblastic tumor (IMT) cases.

| Authors             | Age (years) / Sex | Manifestations              | Laboratory data | Location                        | Size (cm) | Pathological features (Biopsies)       | Treatment              | Follow-up                          |
|---------------------|-------------------|-----------------------------|-----------------|---------------------------------|-----------|----------------------------------------|------------------------|-----------------------------------|
| This patient        | 55/F              | No symptoms                 | Normal          | GC side of middle body          | 1         | Chronic inflammation                   | Surgical resection     | No recurrence in 24 months by PET scan |
| Chow et al. [12]    | 27/M              | Retrosternal pain and dysphagia | Normal          | Gastroesophageal junction       | 7.0 × 4.0 | Chronic inflammation                   | Surgical resection     | No recurrence in 18 months by CT scan |
| Lee et al. [13]     | 42/F              | Epigastralgia and melena    | Anemia          | AW of lower gastric body        | 5.5       | NA                                     | Surgical resection     | NA                                |
| Fong et al. [14]    | 56/M              | Epigastralgia               | Normal          | PW of the gastric body          | 10.0 × 6.0 × 9.0 | NA                                     | Surgical resection     | NA                                |
| Al Hatlani et al. [15] | 11/F            | Weight loss, fatigue        | Anemia          | LC of the stomach               | 4.7 × 3.8 x 3.5 | NA                                     | Surgical resection     | No recurrence in 18 months          |
| Shah et al. [16]    | 80/F              | Epigastric discomfort       | Anemia          | Prepyloric region               | 1.5       | Erosive chronic gastritis and reactive epithelial-cell atypia | Endoscopic mucosal resection (EMR) | NA                                |
| Authors                | Age/ Gender | Symptoms                                      | Findings                                      | Treatment                      | Outcome                  |
|------------------------|-------------|-----------------------------------------------|-----------------------------------------------|-------------------------------|--------------------------|
| Qiu et al. [17]        | 61/F        | High fever with abdominal pain                | Anemia                                        | LC of the antrum              | 3.0 x 3.0                | Surgical resection       | No recurrence in 3 months |
| Jain et al. [18]       | 35/F        | Fever with abdominal pain                     | Anemia                                        | GC of gastric body, and abutting the pancreas | 11.0 x 8.0 x 7.0          | NA                       | Surgical resection       | NA                        |
| Park et al. [19]       | 55/F        | Abdominal pain                                | Anemia, elevated ESR                         | LUQ mass and attached to GC of the stomach | 8.5 x 7.1 x 3.6            | NA                       | Surgical resection       | No recurrence when follow-up |
| Arslan et al. [20]     | 65/F        | Dyspepsia, epigastralgia                      | NA                                            | Antrum                        | 7.5 x 10.0 x 11.0         | NA                       | Surgical resection       | No recurrence in 37 months |
| Bjelovic et al. [21]   | 43/F        | Epigastralgia, nausea and pyrosis             | NA                                            | LC side near angle            | 2.5 x 1.7                | Unclear hypercellular proliferation and inflammatory cells | Surgical resection       | No recurrence in 24 months |
| Kojimahara et al. [22] | 19/F        | Vomiting and weight loss                      | NA                                            | Beneath the cardiac region    | 9                        | NA                       | Surgical resection       | No recurrence in 30 months |
| Kim et al. [6]         | 26/M        | Abdominal mass                                | Normal                                        | From lower Esophagus to gastric body | 8                        | NA                       | Surgical resection       | F/u 5 weeks with recurrence in the rectovesical pouch |
| Leon et al. [23]       | 50/F        | Vomiting and weight loss                      | Normal                                        | Near Gastrojejunostomy        | 7                        | Necrotic granulation tissue | Surgical resection       | No recurrence in 24 months |
| Albayrak et al. [24]   | 56/M        | Hematemesis and melena                        | Anemia, leukocytosis                          | From the cardia and extending towards the pylorus | 6                        | Normal mucosa            | Surgical resection       | No recurrence in 8 months |
| Ribeiro et al. [25]    | 37/F        | Weight loss                                   | Anemia                                        | PW of lower gastric body      | 9.0 x 7.0 x 6.0           | NA                       | Surgical resection       | No recurrence in 5 months |
| Hirschburger et al. [26]| 8mon/F     | Vomiting                                      | Normal                                        | Bursa omentalis close to the gastric antrum | 8.0 x 6.0 x 7.0           | NA                       | Surgical resection       | No recurrence in 36 months |
| Cho et al. [27]        | 2/M         | Fever, poor appetite                          | Anemia                                        | GC of fundus                  | 9.0 x 7.0 x 6.0           | Spindle cells and some atypical cells with a high N/C ratio | Surgical resection       | No recurrence in 3 months |
| Riedel et al. [28]     | 4mon/F      | Hematemesis                                   | Anemia                                        | From gastric cardia to gastric wall and spleen | 4                        | NA                       | Surgical resection       | No recurrence in 24 months |
| Marves et al. [29]     | 5/F         | Melena                                        | Anemia, Elevated IgA                          | Gastric fundus                | 6.0 x 8.0                | NA                       | Surgical resection       | No recurrence in 48 months |
| Marves et al. [29]     | 18mon/F     | Fever and weight loss with LUQ mass           | Anemia                                        | Mass adherent to GC of stomach | NA                      | NA                       | Surgical resection       | No recurrence in 11 months |
| Taratuta et al. [30]   | 5/F         | Anorexia and vomiting                         | Leukocytosis, Anemia, Elevated ESR, Liver function, LDH | From LC of stomach to omentum | 7.0 x 6.0 x 5.0           | NA                       | Surgical resection       | No recurrence in 4 months |
| Kim et al. [31]        | 5/F         | Vomiting and weight loss                      | Normal                                        | From GE junction to LC of stomach | NA                      | NA                       | Surgical resection       | No recurrence in 8 months |
| Hoseini-Azar et al. [32]| 18/M       | Nausea, weight loss, epigastric pain, fever    | Anemia                                        | From fundus with extension to the GC side | 10                      | NA                       | Surgical resection       | No recurrence in 9 months |
| Chen et al. [33]       | 50/F        | Abdominal distension                          | Anemia                                        | From the GC of stomach to upper spleen | 22.0 x 13.0 x 8.5         | NA                       | Surgical resection       | No recurrence in 4 months |
| Fragoso et al. [34]    | 10/F        | Gastrointestinal bleeding                     | Anemia                                        | Prepyloric region             | 3                        | NA                       | Surgical resection       | No recurrence in 15 years |
| Fan et al. [35] | 37/M | Abdominal pain | Elevated neuron-specific enolase | Gastric antrum along the LC side | 4.5 × 3.5 | NA | Surgical resection | No recurrence in 6 months |
| Shin et al. [36] | 16/F | Abdominal pain | Normal | Distal body of the stomach | 4.0 × 3.5 | 2.5 | NA | Surgical resection | No recurrence in 3 years |
| Ning et al. [37] | 50/F | Epigastric pain | Normal | Gastric antrum | 3 | NA | Endoscopic submucosal dissection (ESD) | No recurrence in 2 years |
| Junanee et al. [38] | 9/F | Fever, weight loss, hematemesis, melena | Anemia | GC of the stomach infiltrating to the spleen | 8.0 × 10.0 | NA | Surgical resection | No recurrence in 6 months |
| Lazure et al. [7] | 12/M | Abdominal pain, weight loss | Anemia, elevated ESR | PW of the gastric body to spleen | 8 | Suspect IMT | Surgical resection | No recurrence in 4 years |
| Lazure et al. [7] | 11/M | Scapular pain | Anemia | Stomach to esophagus and left pulmonary hilum | 12 | Suspect IMT | Surgical resection + chemotherapy | Recurrence and died 11 months after diagnosis |
| JadHAV et al. [39] | 18/M | Poor appetite, weight loss | Anemia, elevated ESR | LC of the stomach | 9.0 × 9.0 | 7.0 | Suspect GIST or leiomyoma | Surgical resection | No recurrence in 5 years |
| Kim et al. [40] | 28/M | Hematemesis | Anemia | PW of the fundus | 3.6 | | Spindle cell-type lesion | Surgical resection | No recurrence in 18 months |
| Noh et al. [41] | 58/F | No symptoms | Normal | PW near the high gastric body | 2.5 × 2.2 | 1.5 | A type-undetermined mesenchymal tumor | Surgical resection | No recurrence in 3 months |
| Shi et al. [42] | 36/M | Abdominal pain, abdominal mass | NA | LC of gastric antrum | 4.5 | NA | Surgical resection | No recurrence in 5 years |
| Shi et al. [42] | 42/M | Abdominal pain, hematemesis and abdominal mass | NA | GC of gastric body | 8 | NA | Surgical resection (recurrence and received 2nd operation) | No recurrence after received 2nd operation for 2 years |
| Shi et al. [42] | 40/M | Abdominal mass | NA | AW of gastric body | 6.3 | NA | Surgical resection | No recurrence in 3.3 years |
| Shi et al. [42] | 45/M | Abdominal pain and abdominal mass | NA | Gastric angle | 5.5 | NA | Surgical resection | No recurrence in 2.6 years |
| Shi et al. [42] | 40/F | Abdominal pain and abdominal mass | NA | PW of gastric body | 5.8 | NA | Surgical resection | No recurrence in 4 years |
| KataKWAR et al. [43] | 45/M | Weight loss and epigastric pain | Anemia | PW of gastric body | 5.7 × 4.7 | NA | Surgical resection | No recurrence when follow-up |
| Lee et al. [44] | 5/F | Epigastric discomfort and vomiting | NA | Gastric fundus and body | 4.0 × 3.0 | NA | Surgical resection | No recurrence when follow-up |

Table 2: Summarized clinicopathological features from the reviewed literature.

| Parameter                                      | Distribution of patients                                      |
|------------------------------------------------|--------------------------------------------------------------|
| Age                                            | 4 months – 80 years (mean ± SD: 29.83 ± 21.61)               |
| Female: Male                                   | 26:16                                                        |
| Symptoms                                       | n (%)                                                        |
| Abdominal pain and epigastralgia              | 21 (50)                                                      |
| Weight loss                                    | 11 (26.19)                                                   |
| Hematemesis and melena                         | 10 (23.80)                                                   |
| Nausea and vomiting                            | 8 (19.04)                                                    |
| Symptom                  | Count (Percentage) |
|-------------------------|--------------------|
| Abdominal mass          | 7 (16.66)          |
| Fever                   | 6 (14.28)          |
| Poor appetite           | 2 (4.76)           |
| Dyspepsia and pyrosis   | 2 (4.76)           |
| No symptoms             | 2 (4.76)           |
| Dysphagia               | 1 (2.38)           |
| Fatigue                 | 1 (2.38)           |
| Anorexia                | 1 (2.38)           |
| Abdominal distension    | 1 (2.38)           |
| Scapular pain           | 1 (2.38)           |

| Laboratory Data          | Count (Percentage) |
|--------------------------|--------------------|
| Anemia                   | 22 (66.66)         |
| Normal                   | 10 (30.30)         |
| Elevated ESR            | 4 (12.12)          |
| Leukocytosis             | 2 (6.06)           |
| Elevated IgA             | 1 (3.03)           |
| Impaired liver function  | 1 (3.03)           |
| Elevated LDH             | 1 (3.03)           |
| Elevated neuron-specific enolase | 1 (3.03) |
| Tumor size: Range       | 1.0-13.0           |

| Location                 | Count (Percentage) |
|--------------------------|--------------------|
| Body                     | 18 (42.85)         |
| Antrum                   | 6 (14.28)          |
| LC side of stomach       | 5 (11.90)          |
| GC side of stomach       | 5 (11.90)          |
| Cardia                   | 3 (7.14)           |
| Gastroesophageal junction| 2 (4.76)           |
| Pyloric region           | 2 (4.76)           |
| Angle                    | 1 (2.38)           |
| Gastrojejunostomy        | 1 (2.38)           |
| Stomach to left pulmonary hilum | 1 (2.38) |

| Biopsied Specimen        | Count (Percentage) |
|--------------------------|--------------------|
| Chronic inflammation, inflammatory cell, reactive gastritis | 5 (38.46) |
| Spindle cell type lesion | 2 (15.38)          |
| Suspect IMT              | 2 (15.38)          |
| Necrotic granulation tissue | 1 (7.69)       |
| Normal mucosa            | 1 (7.69)           |
| Suspect GIST or leiomyoma| 1 (7.69)           |
| Undetermined mesenchymal tumor | 1 (7.69) |

| Treatment                | Count (Percentage) |
|--------------------------|--------------------|
| Surgical resection       | 39 (92.8)          |
| Surgical resection + chemotherapy | 1 (2.38) |
| EMR                      | 1 (2.38)           |
| ESD                      | 1 (2.38)           |
| Follow-up                | 3 months – 5 years |
| No recurrence            | 35 (92.10)         |
| Recurrence               | 3 (7.89)           |
Hypoechoic mass
Subepithelial layer
Hypoechoic lesion
Muscularis propria
Hypoechoic lesion
Heterogeneous and Solid protruding mass
Submucosal layer
Origin of Layer
Muscularis propria
Multicystic lesion
Muscularis propria
Summarized EUS finding.

NA (origin from Table 3) (the features of IMT by EUS. The most common IMT origin layer is the muscularis propria layer (7/39). The summarized clinical features are shown in Table 2.

Radiological Diagnosis

There is no radiological sign that serves as a diagnostic basis for gastric IMT [40-45]. Gastric IMTs have been reported to present as heterogeneous, lobular, calcified, cystic lesions on ultrasound and non-enhanced abdominal CT examinations [1], though they may manifest as homogeneous or heterogeneous lesions with central delayed enhancement and peripheral early filling [45,46]. Abdominal CT may help to determine the invasion of gastric IMTs to adjacent organs [6].

Endoscopic Diagnosis

The diagnostic role of Endoscopic Ultrasound (EUS) remains unclear in previous clinical settings. From the reviewed literature, EUS was only performed in a few patients. We identified patients who underwent EUS, and they are summarized in Table 3. Ten patients in the reviewed literature underwent EUS before surgical treatment. The most common feature in the EUS results was a hypoechoic mass (5/10). Other features such as a hyperechoic, heterogeneous lesion were also reported in the reviewed literature. The most common IMT origin layer is the muscularis propria layer (7/10). The submucosal and subepithelial layers were also reported in the literature. The differential diagnosis of submucosal tumors arising from the muscularis propria layer included Gastrointestinal Stromal Tumor (GIST), leiomyoma, and schwannoma. Hypoechoic lesions observed through echogenicity were found in these submucosal tumors. According to the most common EUS results for IMT in the reviewed literature, IMT with a hypoechoic lesion arising from the muscularis propria layer might cause a differential diagnosis. To our best knowledge, this is the first article to discuss the features of IMT by EUS.

Table 3: Summarized EUS finding.

| Author            | EUS Finding                          |
|-------------------|--------------------------------------|
|                   | Features                             | Origin of Layer |
| This patient      | Focal thickening lesion               | Muscularis propria layer |

PET Diagnosis

In our patient, the diagnosis was based on positron emission tomography. Dong et al. has previously reported a mean SUVmax of 10.9±5.5 for IMT, ranging from 3.3 to 20.8 [47]. The wide variability, which is believed to be attributed to varying proportions of inflammatory cells in the tumor [47], renders PET suboptimal for differentiating IMTs from other tumors. By contrast, PET appears to be a useful tool for the follow-up of IMT relapse after treatment, which is typically reflected by an elevation of SUVmax. In our case, the SUVmax of the gastric IMT was 6.21. The four factors that have been found to affect the tissue uptake of fluorodeoxyglucose by PET include tumor cellularity, the biological behaviors of tumor cells, the composition and proportion of inflammatory cells, and the degree of inflammatory cell activation [47].

Histological Features

The typical histological features of IMT include myofibroblastic proliferation with lymphoplasmacytic infiltration in myxoid background stroma [48]. IMT is known to have the potential for malignant transformation and metastasis. Recurrence, which is not uncommon for incompletely resected lesions, has been reported to occur in 7% (3 in 38) of patients with a mortality rate of 5% (2 in 38) [49]. Local recurrence of IMT has been shown to be related to the rearrangement of the anaplastic lymphoma kinase (ALK) gene on chromosome 2p23 [4]. Indeed, chromosomal translocations of active ALK gene have been found in almost 50% of IMTs. Previous studies have indicated that ALK gene translocation, which may be associated with a higher recurrence rate, mostly occurs in children and young adults. By contrast, IMTs without an ALK gene translocation are mostly found in older people and are associated with distal metastasis. Moreover, cellular atypia, a ganglion-like cell morphology, aneuploidy, and overexpression of p53 have been reported to be associated with tumor aggressiveness [4,50]. Consistently, our patient showed no evidence of an ALK gene translocation. The positivity of staining for cytokeratin, laminin,
calponin, smooth muscle actin (SMA), muscle-specific actin (MSA), fibronectin, and desmin also varies in IMTs and cannot provide reliable diagnostic clues [12].

**Treatment Strategies and Prognosis**

Complete surgical resection is the most efficient treatment for gastric IMTs. Among the gastric IMT patients, two were treated with endoscopic mucosal resection (EMR) and endoscopic submucosal dissection (ESD), while the rest underwent surgical resection. The indication of EMR or ESD for a particular patient was due to the relatively small size of the tumor together with its favorable location for the endoscopic approach. Incompletely resected IMTs have been reported to have a high one-year local recurrence rate [6,51] with an overall probability of recurrence up to 60% [5,52]. Survival after complete resection is 91% at 5 years and 77% at 10 years [51]. Partial resection may be considered when complete resection is inappropriate for selected patients with severe co-morbidities to whom radiotherapy or chemotherpay (cyclosporine, methotrexate, azathioprine, and cyclophosphamide) may be applied for the relief of symptoms [4,8]. Recent advancements in IMT treatment include the use of an anaplastic lymphoma kinase (ALK) inhibitor, which has been reported in two patients with abdominal and pancreatic IMTs with recurrence after surgical resection, one of whom showed a partial response. The other patient was withdrawn from the trial due to disease progression [53]. The benefit of including an ALK inhibitor as a standard therapeutic agent against IMTs remains to be elucidated.

**Conclusion**

Albeit uncommon, the diagnosis of gastric IMT should not be ruled out upon encountering asymptomatic gastric tumors during endoscopic examination. Gastric IMT might give rise to a differential diagnosis when hypoechoic lesions arise from the muscularis propria layer by EUS. Although the major treatment for gastric IMT is surgical resection, due to the high recurrence rate for incompletely resected tumors, chemotherapy and radiotherapy may be considered for patients with unresectable lesions for symptom relief. Translocation of the ALK gene may play an important role and may be associated with outcomes. An ALK gene inhibitor might provide new treatment options for IMTs with recurrence in the future.

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