A Case of Inflammatory Myofibroblastic Tumor Originating in the Lesser Omentum Treated by Laparoscopic Resection

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Abstract
A 51-year-old woman underwent a CT scan performed that revealed a neoplastic lesion in the upper abdomen, and she was referred to our surgical department. Abdominal CT showed a tumor of about 50 mm lying between the posterior wall of the gastric minor curvature and the pancreas. Continuity with the surrounding tissue was not clear. Ultrasonography showed that the tumor was mobile on postural change, and it was considered to originate from the mesentery. In addition, PET scanning showed mild accumulation, and MRI showed limited diffusion, suggesting the possibility of malignancy. Laparoscopic tumor resection was performed as a diagnostic treatment. Intraoperative findings showed tumor continuity only with the lesser omentum, and the patient was judged to have a primary tumor of the lesser omentum. Histopathological findings showed proliferation of myofibroblasts and lymphocyte infiltration, and the patient was thus diagnosed as having an inflammatory myofibroblastic tumor. Inflammatory myofibroblastic tumor as a primary tumor of the lesser omentum is rare and is reported here along with a literature review.

Key words
Inflammatory myofibroblastic tumor, laparoscopic surgery, lesser omentum

Introduction
Inflammatory myofibroblastic tumors (IMTs) are neoplastic lesions that are characterized by the proliferation of myofibroblasts and significant inflammatory cell infiltrate. This report documents a case of IMT originating in the lesser omentum that we encountered and resected laparoscopically, along with a review of the relevant literature.

Case Presentation
The patient was a 51-year-old woman with no chief complaints. She had a medical history of hyperlipidemia and cholecystolithiasis but no family history of note. The patient noticed a tumor in her right breast and was examined by our hospital, where she was diagnosed as having right breast cancer. A computed tomography (CT) scan performed for detailed investigation revealed a neoplastic lesion in the epigastric region, so she was referred to our surgical department.

At the time of admission, her vital signs were height, 152 cm; weight, 53 kg; blood pressure, 119/80 mmHg; pulse, 70 bpm; body temperature, 36.2°C. Her palpebral and scleral conjunctiva showed no signs of anemia or jaundice. There were no palpable superficial lymph nodes. During examination of the abdomen, we did not observe any findings suggesting peritoneal irritation, such as tenderness or rebound tenderness. There was no obvious, palpable mass, and the patient did not exhibit hepatosplenomegaly.

Blood test findings did not show any specific abnormal findings (Table 1). An abdominal ultrasound

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showed a richly perfused tumor measuring 58 × 46 mm in size between the stomach and tail of the pancreas. The tumor position changed when the patient was moved into an upright position, so we believed that the tumor was of mesenteric origin. A contrast-enhanced CT scan showed a partially calcified tumor with distinct margins measuring 42 × 38 mm near the posterior gastric wall along the lesser curvature. There was intense contrast enhancement during the early phase. The degree of continuity with the surrounding organs was unclear, but the findings suggested that the tumor could be of gastric origin (Fig. 1).

An esophagogastroduodenoscopy revealed a

| Table 1. Preoperative Test Results |
| WBC  | 5.0 x10^3/μl | LDH  | 157 U/l | Insulin | 1.9 μU/ml |
| Hb  | 13.6 g/dl    | ALP  | 320 U/l | Glucagon | 100 pg/ml |
| Plt | 129 x10^3/μl | γ-GTP| 32 U/l | Gastrin  | 90 pg/ml |
|     |             | BUN  | 11.4 mg/dl |        |          |
| PT-INR | 1.02 | Cre  | 0.60 mg/dl CA15-3 | 5.3 U/ml |
| APTT | 37.4 sec ↑ | Na  | 140 mEq/l | CEA | <0.5 ng/ml |
|      |        | K   | 4.1 mEq/l CA19-9 | 8.1 U/ml |
| TP  | 7.2 g/dl   | Amy | 49 U/l | Elastase-1 | 46 ng/dl |
| Alb | 4.2 g/dl   | CRP | <0.03 mg/dl DUPAN-2 | <25 U/ml |
| AST | 15 U/l    |     | Span-l |          | 7 U/ml |
| ALT | 13 U/l    | HbA1c | 5.6 % |        |          |

Fig. 1. On CT imaging, a smooth tumor with calcification measuring 45 × 38 mm was observed close to the stomach. The tumor was enhanced strongly in the arterial phase. Because the border between the tumor and stomach was obscure, the tumor was thought to have invaded the stomach. (i) Arterial phase. (ii) Venous phase. (iii) Coronal plane. (iv) Sagittal plane.
fundus gland polyp, but no other elevated lesions were observed. Magnetic resonance imaging (MRI) findings revealed a tumor appearing hypointense on T1 images and mildly hyperintense on T2 images that was not continuous with either the pancreas or stomach. Diffusion-weighted images suggested the possibility of malignancy due to restriction of diffusion. Positron emission tomography-CT (PET-CT) scanning showed that the tumor exhibited mildly increased uptake with a maximum standardized uptake value (SUVmax) of 3.5.

Although the above-mentioned findings did not allow us to reach a diagnosis, we believed that this tumor was of mesenteric origin and that the potential for malignancy could not be ruled out. Leiomyosarcoma, schwannoma, and GIST were considered as differential diagnoses. We thus performed laparoscopic tumor resection for the purpose of diagnosis and treatment and planned to perform surgery for the right breast cancer thereafter.

**Surgical findings:** The surgery was performed laparoscopically under general anesthesia with the patient in the supine position. We inserted a total of three ports: a 10-mm umbilical camera port, and two 5-mm ports in the left flank and in the left hypogastrium below the midline. When we opened the omental bursa, we observed a red tumor with a smooth surface. The tumor was not continuous with either the pancreas or stomach and only exhibited continuity with the lesser omentum, so we determined that it was of omental origin (Fig. 2 A, B). We then resected the tumor. Surgery lasted 160 min, and the volume of blood loss was 70 ml.

**Resected specimen findings:** Macroscopic examination of the resected specimen revealed a partially calcified solid tumor measuring 55 × 45 × 25 mm. The histological examination revealed atypical, fusiform, oval-shaped cells, and immunological staining revealed increased proliferation of α-smooth muscle actin (αSMA)-positive myofibroblasts and extensive interstitial infiltration by non-atypical lymphocytes. We therefore diagnosed the patient as having IMT. The MIB-1 index was <5%. In addition, the patient tested negative for anaplastic lymphoma kinase-1 (ALK), which is thought to be a highly specific test for IMT (Fig. 3 A, B).

**Postoperative clinical course:** The patient’s postoperative course was favorable, and she was discharged on postoperative day 5. To date, 4 years and 5 months after the surgery, no recurrence of findings has been observed.

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**Fig. 2.** Port placement and intraoperative images. A Port placement. B- (i) The tumor was covered with greater omentum. (ii) On opening the omental bursa, we observed a smooth red tumor. Because it continued not with the stomach and pancreas but with the lesser omentum, we considered that the tumor had arisen from the lesser omentum. (iii) We separated the tumor from lesser omentum. (iv) We resected the tumor.
Fig. 3. Macroscopic and microscopic findings of the tumor. A. Macroscopically, the tumor was solitary and measured 55 × 45 × 25 mm. B. (i) Pathology findings showed the tumor cells to be in a deformed spindle or oval shape, and many lymphocytes had invaded the tumor. The yellow arrows indicate the spindle tumor cells. (ii) The tumor was positive for α-SMA. (iii) The tumor was negative for ALK. (iv) The MIB-1 index was <5%.

Discussion

IMT develops as a result of myofibroblast proliferation, and these tumors are characterized by the presence of myofibroblasts and extensive lymphocytic infiltrates\(^1\). Although numerous causes have been proposed, such as infection or inflammation as a result of trauma, or surgery or malignant tumors, their origin remains unknown. According to reports, the lungs are the most common site of onset, although IMTs may arise throughout the body, including the peritoneum, mesentery, and bowel. Onset has also been reported in the bladder, endocrine organs such as the prostate, and in the breast, spleen, pancreas, parenchymal intra-abdominal organs such as the liver, and even in the soft tissues\(^2,3\). The condition tends to occur from childhood to young adulthood\(^4\). Symptoms vary based on the site of onset and tumor diameter. However, as in our case, a few reports state that the tumor may be asymptomatic, but the patient may present with pyrexia or abdominal pain. Imaging is not adequately specific for diagnosis, so the diagnosis is based on histopathological tests\(^4\). Differenti-
ating diseases of primary IMT in the abdominal cavity include schwannoma, GIST, and leiomyosarcoma. It is important to identify the primary organ for differentiation, and this can be judged comprehensively by combining CT, MRI, and endoscopy. In addition, PET may be used to judge benign/malignant tumors, but IMT may show faint accumulation on PET, as in our case, which is a reference-level finding. None of these diseases have specific diagnostic imaging characteristics, and it is considered difficult to distinguish them. IMT may present with the following three histological patterns: 1) a myxoid/vascular pattern, with scant distribution of spindle-shaped cells accompanied by an inflammatory cell infiltrate, 2) a compact spindle cell pattern, with a dense arrangement of hyperproliferative spindle-shaped cells, and 3) a hypocellular fibrous pattern, with scant spindle-shaped cells in a collagen fiber matrix resembling scar tissue, accompanied by an inflammatory cell infiltrate, and occasionally associated with calcification or ossification. We believe that we observed pattern 1 in the present case.

Immunologically, myofibroblast cell lines tend to test positive for αSMA, vimentin, and desmin at higher rates than normal. Characteristically, these cell lines are also said to test positive for chromosomal anaplastic lymphoma kinase (ALK) at a higher rate. Our patient tested positive for αSMA but negative for ALK. The ALK inhibitor crizotinib has been shown to be effective for ALK-positive IMT, whereas testing negative for ALK has also been reported to be a factor indicating a poor prognosis for this condition.

When we searched PubMed using the keywords “omentum” and “inflammatory myofibroblastic tumor”, we found 12 reported cases of onset in the greater or lesser omentum, including the present case (we excluded cases in which we were unable to obtain the full report, or in which information was insufficient). Table 2 summarizes our case and the 11 other reported cases. The median age of onset was 20 years, and the initial symptom was abdominal pain in seven cases, a feeling of abdominal discomfort in one case, awareness of an abdominal mass in three cases, and one case was asymptomatic. There were various imaging findings, although findings sug-

Table 2. Review of 12 Cases of IMT Originating in the Omentum

<table>
<thead>
<tr>
<th>Reference</th>
<th>Year</th>
<th>Age</th>
<th>Sex</th>
<th>Initial complaint</th>
<th>Location</th>
<th>Tumor size (mm)</th>
<th>Treatment</th>
<th>ALK</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gupta</td>
<td>2009</td>
<td>6</td>
<td>M</td>
<td>Abdominal pain</td>
<td>Greater omentum</td>
<td>55x36</td>
<td>Resection (laparotomy)</td>
<td>N.D.</td>
</tr>
<tr>
<td>Uchiyama</td>
<td>2009</td>
<td>24</td>
<td>M</td>
<td>Abdominal pain</td>
<td>Lesser omentum</td>
<td>99x55x30</td>
<td>Resection (laparotomy)</td>
<td>-</td>
</tr>
<tr>
<td>Sodhi</td>
<td>2010</td>
<td>2</td>
<td>M</td>
<td>Abdominal lump</td>
<td>Greater omentum</td>
<td>150x100x90</td>
<td>Resection (laparotomy)</td>
<td>N.D.</td>
</tr>
<tr>
<td>Singhal</td>
<td>2011</td>
<td>15</td>
<td>F</td>
<td>Abdominal pain</td>
<td>Greater omentum</td>
<td>72x63</td>
<td>Resection (laparotomy)</td>
<td>+</td>
</tr>
<tr>
<td>Kye</td>
<td>2012</td>
<td>22</td>
<td>F</td>
<td>Low abdominal mass</td>
<td>Greater omentum</td>
<td>75x70x50</td>
<td>Resection (laparotomy)</td>
<td>+</td>
</tr>
<tr>
<td>Aptel</td>
<td>2012</td>
<td>20</td>
<td>F</td>
<td>Hypogastric mass</td>
<td>Greater omentum</td>
<td>70</td>
<td>Resection</td>
<td>+</td>
</tr>
<tr>
<td>Yagmur</td>
<td>2014</td>
<td>53</td>
<td>F</td>
<td>Abdominal pain</td>
<td>Greater omentum</td>
<td>70x60</td>
<td>Resection (laparotomy)</td>
<td>+</td>
</tr>
<tr>
<td>Cianci</td>
<td>2015</td>
<td>75</td>
<td>M</td>
<td>Abdominal pain</td>
<td>Greater omentum</td>
<td>260</td>
<td>Resection (laparotomy)</td>
<td>-</td>
</tr>
<tr>
<td>El Hage</td>
<td>2016</td>
<td>38</td>
<td>M</td>
<td>Lower abdominal pain</td>
<td>Greater omentum</td>
<td>70</td>
<td>Resection (laparotomy)</td>
<td>+</td>
</tr>
<tr>
<td>Oeconomopoulou</td>
<td>2016</td>
<td>6</td>
<td>M</td>
<td>Abdominal pain</td>
<td>Greater omentum</td>
<td>60x24</td>
<td>Resection (laparoscopy)</td>
<td>-</td>
</tr>
<tr>
<td>Liang</td>
<td>2017</td>
<td>25</td>
<td>F</td>
<td>Abdominal discomfort</td>
<td>Greater omentum</td>
<td>60x60x70</td>
<td>Resection (laparoscopy)</td>
<td>+</td>
</tr>
<tr>
<td>Our case</td>
<td>2017</td>
<td>51</td>
<td>F</td>
<td>None</td>
<td>Lesser omentum</td>
<td>55x45x25</td>
<td>Resection (laparoscopy)</td>
<td>-</td>
</tr>
</tbody>
</table>

N.D., not described.
gestive of IMT were observed on PET in certain cases, and these findings were difficult to differentiate from malignancy in some cases. Surgical resection was performed in all cases, although including our case, laparoscopic resection was performed in only three cases. We believe that the facts that the tumors were comparatively large at the time of detection and that there were numerous young patients may have contributed to this. The results for immunostaining showed that six patients tested positive for the aforementioned ALK, whereas four tested negative, and the outcome in two patients was unknown. Therefore, approximately half of all cases tested positive for ALK. Reports indicate that between 5 and 60% of patients test positive for ALK, and the rate of positive tests reportedly varies based on the site of onset. The results in the present tabulation resemble those in previous reports. It would be desirable to accumulate further cases going forward.

According to the World Health Organization (WHO) classification, IMT is an intermediate (rarely metastasizing) tumor. Originally, the observation of a mixture of proliferating fibroblasts, myofibroblasts, and inflammatory cells was associated with inflammatory changes, such as granulation tissue, so previously, these lesions were understood to be reactive lesions and were referred to in general terms, such as plasma cell granulomas and inflammatory pseudotumors. However, in the interim, there have been cases in which these tumors exhibited local recurrence, distant metastasis, and infiltration, so they are now defined as intermediate tumors. Cases of local recurrence usually occur as a result of inadequate resection or careless biopsy, so generally, complete resection is considered to be the appropriate form of treatment. However, when we consider the fact that IMT onset may be caused by invasive surgical procedures, we believe that it is desirable to perform minimally invasive surgery. If elective surgery is permissible, then we believe that laparoscopic surgery is a useful technique that will allow both examination and diagnosis and minimally invasive treatment in comparison with open resection.

Conclusion

We experienced a case of IMT originating from the lesser omentum. Although IMT originating from the lesser omentum is extremely rare, it should be kept in mind as a differential disease when evaluating nonspecific intra-abdominal tumors.

Conflicts of Interest

The authors have nothing to disclose.

References

11) El Hage Chehade HH, Zbibo RH, Abou Hussein BM, et al. Highly vascularized primarily inflam-


