A Rare Case of Ileocecal Lymph Node Recurrence After Surgery in Siewert’s Classification Type I Esophagogastric Junction Adenocarcinoma

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Patient: Male, 52-year-old
Final Diagnosis: Solitary ileocecal lymph node recurrence after surgery for esophagogastric junction adenocarcinoma
Symptoms: No symptom
Clinical Procedure: —
Specialty: Gastroenterology and Hepatology • Surgery

Objective: Unusual clinical course
Background: Although recurrence after surgery for esophagogastric junction (EGJ) adenocarcinoma frequently develops in the mediastinal and para-aortic lymph nodes (LN), distant LN recurrence in the mesocolon is rare. We report a rare case of ileocecal LN metastasis in the ascending mesocolon after radical surgery for an EGJ adenocarcinoma.

Case Report: We performed subtotal esophagectomy with mediastinal and para-gastric LN dissection in a patient with an advanced EGJ adenocarcinoma. Clinopathologically, the patient was diagnosed with type I EGJ adenocarcinoma based on Siewert’s classification (pathological T3N1M0). One year after surgery, computed tomography showed enlarged lymph nodes around the ileocolic artery, and further examination was performed. Although positron emission tomography-computed tomography showed that the lesion had moderate uptake of fluorodeoxyglucose, we did not find the reason for the enlarged lymph nodes. Finally, laparoscopic ileocecal resection was performed for diagnostic and therapeutic purposes. Clinicalopathological tests revealed that the specimen was a moderately differentiated adenocarcinoma, which was strongly suspected to be a metastasis of the EGJ adenocarcinoma.

Conclusions: We encountered a rare case of EGJ adenocarcinoma that spread to the ileocecal LN in the ascending mesocolon. To the best of our knowledge, this is the first such report in the literature to date. Laparoscopic ileocecal resection for metastasis to the ascending mesocolon seems reasonable as a local control.

Keywords: Esophagectomy • Adenocarcinoma • Mesocolon • Lymph Nodes • Laparoscopy

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Introduction

Esophageal cancer remains a refractory malignancy because of high recurrence of disease after curative treatments. It is reported that locoregional recurrence and distant recurrences, including lung, liver, bones, and brain, develop in 22-68% and 12-51% of cases, respectively [1].

Recently, the incidence of esophagogastric junction (EGJ) adenocarcinoma has gradually increased in Japan. Risk factors for EGJ adenocarcinoma include the presence of GERD, high body mass index, and smoking. The symptoms of EGJ adenocarcinoma are not as common as those of esophageal cancer in the early stages. However, a mild pinching sensation and symptoms of heartburn after drinking hot beverages may be present. As EGJ adenocarcinoma progresses, esophageal stricture occurs, resulting in decreased food intake and weight loss.

In Siewert's classification, EGJ is categorized into types I to III. While in Western countries these types exist in roughly equal proportions, in Japan, type I accounts for less than 5%, with type II and type III comprising equal proportions [2]. Standard therapy consists of radical surgery and lymph node dissection. Although the extent of dissection varies by type, the approach to standard therapy remains unchanged. Chemotherapy is administered either before or after surgery, depending on the stage of the disease, but the superiority of preoperative or postoperative chemotherapy has not been established. In cases of advanced, unresectable disease, the 5-year survival rate is reported to be less than 30% [2].

Although radical surgery with lymph node (LN) dissection is performed for advanced EGJ adenocarcinoma, the incidence of recurrence after surgery is relatively high [3]. The recurrence pattern depends on the Siewert classification subtype. The LN was the most frequent relapse site of EGJ adenocarcinoma type I, although hematogenous and disseminative recurrences were found in types II and III, respectively [4]. The most common sites of LN recurrence are the para-aortic and mediastinal regions. Solitary recurrence in extra-regional LNs is extremely rare. Herein, we report a rare case of solitary ileocecal LN recurrence in the ascending mesocolon after surgery for EGJ adenocarcinoma.

Case Report

The patient was a 52-year-old man who had undergone an upper gastrointestinal endoscopy for cancer screening. He was referred to a large academic medical center in Tokyo, Japan because of advanced cancer just above the EGJ incidentally found in the cancer screening. He had no subjective symptoms, and his medical history included colon polyps, ventricular tachycardia, and diabetes mellitus, but no history of malignancy. We planned treatment for an advanced EGJ adenocarcinoma. We first conducted 2 courses of preoperative chemotherapy, consisting of fluorouracil (1800 mg, intravenous, days 1-5, followed by a 3-week rest period) plus cisplatin (185 mg, intravenous, day 1), and then performed subtotal esophagectomy for an advanced EGJ adenocarcinoma. Lymphadenectomy was performed on the mediastinal and para-gastric LNs, including #1-3, 7, 8a, 105-110 and 112 LNs, which were classified according to the Japanese Classification of Esophageal Cancer [5].

Figure 1. Tumor of the esophagogastric junction. Gastroscopic image (A). The resected specimen. The tumor was located 2 cm from the cardia and was diagnosed as type 1 according to the Siewert classification (B).
Clinicopathologically, the patient was diagnosed with Siewert classification type I EGJ adenocarcinoma, and the TNM stage was pathological T3N1M0 in the Japanese Classification of Carcinoma (Figure 1). There was also 1 lymph node metastasis: #7LNs along the left gastric artery. The patient received 8 courses of postoperative chemotherapy, consisting of S-1 (60 mg, orally, days 1-14, followed by a 1-week rest period) plus oxaliplatin on day 1 in a 3-week schedule. The patient was followed up regularly as an outpatient, with blood samples and computed tomography scan at 4-month intervals.

Computed tomography scan 1 year after surgery showed enlarged LNs around the ileocolic artery, and we conducted a detailed examination to make a diagnosis. Preoperative tumor markers were not significantly elevated: carcinoembryonic antigen (CEA), squamous cell carcinoma-related antigen (SCC), and anti-p53 antibody was 1.2 ng/mL (reference value ≤5 ng/mL), 1.01 ng/mL (reference value ≤1.5 ng/mL), and 1.18 U/mL (reference value ≤1.3 U/mL), respectively. Postoperative tumor markers were slightly elevated (SCC: 2.89 ng/mL, anti-p53 antibody: 1.38 U/mL), and positron emission tomography-computed tomography (PET-CT) imaging revealed no findings in the whole body, excluding enlarged LNs around the ileocolic artery, which had moderate uptake of fluorodeoxyglucose (SUV max: 3.19). In addition, colonoscopy did not reveal a cause for the LN swelling in the mesocolon (Figure 2). Therefore, we performed laparoscopic ileocecal resection with complete mesocolic excision for diagnostic and therapeutic purposes (Figure 3) [3]. No complications occurred postoperatively. Postoperative clinicopathological examination of the dissected LNs showed fusion of atypical cells with a rounded enlarged nucleus and basophilic sporangia, as well as formation and proliferation of irregular glandular duct structures. The diagnosis was intermediate-differentiated adenocarcinoma, very similar to the

Figure 2. Computed tomography (CT) and positron emission tomography-computed tomography (PET-CT) scans 1 year after surgery. A solitary lymph node swelling was observed around the ileocolic artery on CT (A, B). Moderate fluorodeoxyglucose (FDG) uptake in the lymph nodes around the ileocolic artery can be observed on the FDG PET-CT image (C, D).
histologic findings of the resected EGJ adenocarcinoma and consistent with metastasis. No tumors were observed in the resected colon (Figure 4). Postoperatively, the patient was followed up without any adjuvant therapy. There was no recurrence of the EGJ adenocarcinoma at 2 years 7 months after ileocelecal resection.

Figure 3. Laparoscopic surgery was performed for diagnostic and therapeutic purposes. A swollen lymph node was found along the ileocolic artery (A). An intracorporeal anastomosis was performed (B). No lesion was found in the resected colon that seemed to cause the swollen lymph node (C).

Discussion

We describe a rare case of distant metastasis to the ileocecal LN in the ascending mesocolon after surgical resection of Siewert’s classification type I EGJ adenocarcinoma.

In the review reported by Shaheen et al [6], esophageal cancer metachronous metastases developed in 58% of patients. Unexpected sites of metastases were found most frequently in the lower one-third of the esophagus, and distal metastases to the gastrointestinal tract are rare. As for EGJ adenocarcinoma, Takeda et al [7] reported recurrence in 72 (38.3%) of 118 patients who underwent curative surgical treatment for

Figure 4. Histologically, the swollen lymph node was finally diagnosed as a lymph node metastasis of an adenocarcinoma of the esophagogastric junction (A: adenocarcinoma of the esophagogastric junction, B: adenocarcinoma in the resected lymph node).
Table 1. Case reports of esophageal cancer distant metastases into colorectal field.

<table>
<thead>
<tr>
<th>Authors</th>
<th>Published year</th>
<th>Metastatic site</th>
<th>Synchronous/metachronous</th>
<th>Treatment</th>
<th>Histological type</th>
<th>Survival</th>
</tr>
</thead>
<tbody>
<tr>
<td>Iwase et al [12]</td>
<td>2004</td>
<td>Sigmoid colon</td>
<td>Synchronous</td>
<td>Chemotherapy</td>
<td>SCC</td>
<td>Died 1 year</td>
</tr>
<tr>
<td>Fang et al [14]</td>
<td>2017</td>
<td>Sigmoid colon</td>
<td>Metachronous</td>
<td>Unknown</td>
<td>SCC</td>
<td>Unknown</td>
</tr>
<tr>
<td>Garg et al [15]</td>
<td>2017</td>
<td>Ascending colon</td>
<td>Metachronous</td>
<td>Radiation</td>
<td>SCC</td>
<td>Died 6 months later</td>
</tr>
<tr>
<td>Wiseman et al [16]</td>
<td>2020</td>
<td>Rectosigmoid</td>
<td>Metachronous</td>
<td>Radiation</td>
<td>SCC</td>
<td>Died 6 months later</td>
</tr>
<tr>
<td>Chen et al [17]</td>
<td>2022</td>
<td>Ascending colon</td>
<td>Synchronous</td>
<td>Unknown</td>
<td>SCC</td>
<td>Unknown</td>
</tr>
<tr>
<td>Zhang et al [18]</td>
<td>2022</td>
<td>Anastomosis of right hemicolecotomy</td>
<td>Synchronous</td>
<td>Chemotherapy</td>
<td>SCC</td>
<td>Unknown</td>
</tr>
<tr>
<td>Makker et al [9]</td>
<td>2016</td>
<td>Rectum</td>
<td>Synchronous</td>
<td>Chemotherapy</td>
<td>AC</td>
<td>Died</td>
</tr>
<tr>
<td>Our case</td>
<td></td>
<td>Ascending mesocolon</td>
<td>Synchronous</td>
<td>Surgery</td>
<td>AC</td>
<td>Alive for 2 years and 7 months</td>
</tr>
</tbody>
</table>

SCC – squamous cell carcinoma; AC – adeno carcinoma. Synchronous: Refers to the situation where multiple tumors are diagnosed at the same time. Metachronous: Refers to the situation where multiple tumors or lesions are diagnosed, with an interval of more than 2 months between the initial and subsequent tumors.

EGJ adenocarcinoma; the most frequent recurrence was distant metastasis, with a rate of 48.6% in 72 patients. Matsuda et al [8] reported that lung recurrence was more common than peritoneal recurrence in 400 patients with Siewert type II EGJ adenocarcinoma. By contrast, Imamura et al [3] reported that 56% of recurrences after surgery for EGJ adenocarcinoma developed in the LNs. The recurrence pattern depends on the Siewert classification subtype. Hosokawa et al [4] reported that the LN was the most frequent relapse site of EGJ adenocarcinoma type I, although hematogenous and disseminative recurrences were found in types II and III, respectively. Colorectal metastasis of EGJ adenocarcinoma is rare. Although we there are reports of colorectal metastasis of esophageal cancer in the PubMed database, all the histological types identified in colorectal metastasis are squamous cell carcinomas except for 1 patient with rectal metastasis reported by Makker et al [9]. Our patient’s case is unique because it describes metastases from esophageal adenocarcinoma rather than squamous cell carcinoma, which is the type of cancer detected in all the other studies except 1. Additionally, we describe ileocecal LN involvement at the ascending mesocolon, which is distinct from the other studies reporting metastases to the ascending colon, transverse colon, sigmoid colon, and rectum (Table 1).

Additionally, we reviewed the literature in Japan over the past 2 decades using the Igaku Chuo Zasshi database, beginning in 2000. We only found 1 case report of an unknown primary carcinoma with ileocecal LN metastasis [10]. Finally, we could not find any reports of patients presenting with distant metastasis to LNs in the ascending mesocolon; thus, our paper is the first such report.

In our patient, the differential diagnoses included gastrointestinal tumors, metastatic tumors, desmoid tumors, and malignant lymphoma. As no tumor causing the swollen LN was visualized on colonoscopy, we performed fluorodeoxyglucose (FDG)-CT to identify the primary tumor. However, moderate FDG uptake was observed only in the LNs around the ileocolic artery. Therefore, we decided to perform laparoscopic surgery for diagnostic and therapeutic purposes, and laparoscopic ileocolic resection with complete mesocolic excision was safely performed. Table 1 shows various treatment options for...
colorectal metastases of esophageal cancer. In general, the prognosis of patients with colorectal metastases is reported to be poor, but our patient has lived without recurrence for 2 years 7 months after surgery.

Conclusions

We encountered a rare case of solitary LN recurrence in the ascending mesocolon after surgery for EGJ adenocarcinoma. Ultimately, we performed laparoscopic surgery for diagnostic and therapeutic purposes with ileocecal resection, with good outcomes. Thus, minimally invasive surgery might be acceptable as a local control therapeutic option for solitary LN recurrence in the ascending mesocolon after surgical resection of type I adenocarcinoma.

References:

1. Guidelines for Diagnosis and Treatment of Carcinoma of the Esophagus, Japan Esophageal Society, Tokyo: Kanehara: 2017