Mass-forming mucosa-associated lymphoid tissue lymphoma of the cecum treated by laparoscopy-assisted bowel resection

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Abstract

Colonic mucosa-associated lymphoid tissue (MALT) lymphoma is very rare, especially those which form a mass. Although the characteristics and treatment methods of gastric MALT lymphomas are well established, those of colonic MALT lymphomas have been insufficiently described. Here, we report a case of mass-forming cecal MALT lymphoma that was successfully treated by laparoscopy-assisted bowel resection. A 60-year-old woman with right lower abdominal pain and a palpable tumor was referred to our hospital. Colonoscopy showed a smooth elevated submucosal tumor-like lesion in the cecum. Histological and immunochemical findings were consistent with MALT lymphoma. Serum tumor marker levels were within normal range. Enhanced abdominal computed tomography showed a large tumor 55 mm in diameter in the cecum and edema of a few paracolic lymph nodes. The tumor was diagnosed as cecal MALT lymphoma classified as stage III1 by Lugano classification, and laparoscopy-assisted ileocecal resection was performed. The postoperative course was uneventful and the patient underwent eradication therapy for *Helicobacter pylori*. A year after the operation she has had no recurrence. In patients with mass-forming colonic MALT lymphoma without dissemination, surgical resection may be a feasible treatment.

Keywords Colonic mucosa-associated lymphoid tissue lymphoma · *Helicobacter pylori* · Laparoscopy-assisted bowel resection

Introduction

Mucosa-associated lymphoid tissue (MALT) lymphoma is a subtype of non-Hodgkin lymphoma and is classified as an extra nodal marginal zone B-cell lymphoma by the World Health Organization [1]. Although most often found in the stomach, 2.5% of MALT lymphomas are found in the colon [2]. In addition, since colonic MALT lymphomas are often detected in the invasion depth of mucosal and submucosal layers as relatively small tumors, they are extremely rarely found as a deeply invasive mass-forming tumor [3–5]. The association between gastric MALT lymphoma and chronic *Helicobacter pylori* (*H. pylori*) infection is evident, so the

treatment of gastric MALT lymphomas is well established. The treatment of colonic MALT lymphomas, however, is not yet standardized. Here, we report a case of mass-forming cecal MALT lymphoma that was successfully treated by laparoscopy-assisted bowel resection.

Case report

A 60-year-old woman was referred to our hospital with right lower abdominal pain and a palpable tumor. She had no remarkable medical, family or social history. Colonoscopy identified a smooth elevated submucosal tumor-like lesion in the cecum (Fig. 1). Tumor biopsy revealed colonic mucosa with dense infiltration of small-to-medium-sized lymphocytes and lympho-epithelial lesion (Fig. 2). Immunohistochemical studies were positive for CD20, and negative for CD3, CD5, CD10 and Bcl-6 (Fig. 3). These findings were consistent with MALT lymphoma. Esophagogastroduodenoscopy identified chronic atrophic gastritis positive for *H. pylori* infection.

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Fig. 1 Colonoscopy showed a smooth elevated submucosal tumor-like lesion in the cecum (**a, b** are the same lesion)

Fig. 2 Biopsy of the tumor revealed colonic mucosa with dense infiltration of small-to-medium-sized lymphocytes and lympho-epithelial lesion (arrow) (H.E. $\times 40$)

Fig. 3 Immunohistochemical studies yielded positive results for CD20 (CD20 $\times 20$)

Fig. 4 Abdominal CT showed a large tumor 55 mm in diameter in the cecum (circle) and a few paracolic lymph nodes' swelling (arrows)

Laboratory evaluation showed slightly low hemoglobin count 11.5 g/dl (normal range 11.6–14.8 g/dl), but other results, including examination for infectious diseases and autoimmune diseases, were unremarkable. Serum tumor marker levels were also in normal range: carcinoembryonic antigen (CEA) in 1.5 ng/ml (normal range ≤ 5.0 ng/ml), carbohydrate antigen 19-9 (CA19-9) in 4.5 U/ml (normal range ≤ 37.0 U/ml) and soluble interleukin-2 (sIL-2) receptor in 581 U/ml (normal range 127–582 U/ml). Further evaluation was performed to assess the stage

Fig. 5 Positron emission tomography revealed high accumulation in the tumor (arrow)

and metastasis of the lymphoma. Enhanced abdominal computed tomography (CT) showed a large tumor in the cecum (diameter 55 mm) and a few paracolic lymph nodes' swelling (≤ 14 mm in diameter) (Fig. 4). Positron emission tomography (PET) revealed high accumulation in the tumor and the lymph nodes, standardized uptake value max was 9.01 and ≤ 2.23 , respectively (Fig. 5). The tumor was thus diagnosed as cecal MALT lymphoma classified as stage II1 according to Lugano classification [6]. As this was a large, localized MALT lymphoma and the patient reported abdominal pain by the tumor, we performed laparoscopy-assisted ileocecal resection with D2 lymphadenectomy. Although adhesion of the tumor to the abdominal wall due to inflammation was observed, the

Fig. 7 Macroscopic findings of the resected specimen revealed a large mass measuring 55 mm $\times$ 30 mm (arrow: cut surface) and its mucosal surface showed some small protruded polypoid lesions

tumor and paracolic lymph nodes were resected in a mass without any complications (Fig. 6). Macroscopic findings of the resected specimen revealed a large mass measuring 55 mm $\times$ 30 mm and its mucosal surface showed some small protruded polypoid lesions (Fig. 7). Histopathologically, diffuse infiltration of atypical lymphocytes from mucosal to subserosal layer was observed and immunohistochemical studies showed the same results as the biopsy (Fig. 8). There was no paracolic lymph node infiltration, and stage I diagnosis was made pathologically according to the classification cited above. The postoperative course was uneventful. After the operation, the patient underwent eradication therapy for *H. pylori* using three administering

Fig. 6 A large mass was located in the cecum and adhesion of the tumor to the abdominal wall due to inflammation was observed

Fig. 8 Microscopic findings showed diffuse infiltration of atypical lymphocytes from mucosal to subserosal layer (**a** H.E. $\times 40$, **b** CD20 $\times 0.75$)

Table 1 Colorectal MALT lymphoma treated by surgical resection in English literature

Case	Author	Year	Age (years)	Sex	Symptom	Location	Tumor size (cm)	<i>H. pylori</i> infection	Surgical method	Pre- or postoperative treatment	Outcome (months)
1	Hasegawa et al. [15]	2000	72	F	Abdominal pain, fever	Sigmoid colon	3.0 $\times$ 1.5	Unknown	Sigmoidectomy	Unknown	Unknown
2	Tanimoto et al. [16]	2002	75	F	Rectal bleeding	Rectum	2.0 $\times$ 1.0 $\times$ 0.7	Unknown	Abdominoperineal resection	Unknown	9, no evidence of disease
3	Takada et al. [14]	2003	44	M	Fecal occult blood	Transverse colon	1.1 $\times$ 0.9	Negative	Laparoscopy-assisted partial resection	Unknown	Unknown
4	Matsuo et al. [4]	2006	75	F	Fecal occult blood	Transverse colon	4.5 $\times$ 3.5	Positive	Partial resection	Preoperative eradication but not responded	12, no evidence of disease
5	Akasaka et al. [3]	2012	50	F	Fecal occult blood	Sigmoid colon	1.8	Unknown	Sigmoidectomy	Unknown	Unknown
6	Jain [17]	2013	55	M	Abdominal pain, distension	Cecum	10.0 $\times$ 5.0	Negative	Right hemicolectomy	None	6, no evidence of disease
7	Seo et al. [18]	2014	64	M	None	Transverse colon	2.5 $\times$ 2.5	Negative	Partial resection	Unknown	12, no evidence of disease
8	Palo and Biligi [19]	2016	68	M	Abdominal pain, bloody stool	Cecum	3.5 $\times$ 2.5 $\times$ 1.8 (multiple)	Unknown	Right hemicolectomy	Unknown	5th day, deceased (surgical complications)
9	Our case	2018	60	F	Abdominal pain	Cecum	5.5 $\times$ 3.0	Positive	Laparoscopy-assisted partial resection	Postoperative eradication	12, no evidence of disease

agents consisting of amoxicillin and clarithromycin antibiotics, and a proton pump inhibitor lansoprazole for 2 weeks, and eradication was confirmed. A year after the operation she has not developed any recurrence.

Discussion

Gastrointestinal lymphomas occur mostly in the stomach and small intestine, much less commonly in the colon and rectum ($\leq 20\%$) [7]. Only 2.5% of all MALT lymphomas are in

the colon [2]. Although the characteristics of gastric MALT lymphomas have been defined and therapeutic strategies have been well studied, optimal treatment for colonic MALT lymphomas remains unclear. The association between gastric MALT lymphoma and chronic *H. pylori* infection is well known, so the standardized therapy for localized gastric MALT lymphoma is eradication of the *H. pylori* [8–10]. Any relationship between colonic MALT lymphoma and *H. pylori* infection, however, is less clear.

Although some reports have described successful regression of colonic MALT lymphoma by *H. pylori* eradication,

other studies, on the contrary, have described its inefficacy [4, 11–13]. *H. pylori* eradication is, therefore, not standardized as an initial therapy in colonic MALT lymphomas. Although various treatment methods including surgery, chemotherapy and radiation have been performed, some reports indicate the effectiveness of surgical resection and suggest it might be the best choice for colonic MALT lymphoma localized without dissemination [4, 14]. Surgical resection has been performed only in limited cases in previous reports, such as those with large tumors or those that were clearly symptomatic. However, they did not develop any recurrence within the described duration (Table 1) [3, 4, 14–19]. Our case was colonic MALT lymphoma localized in the cecum and it was causing abdominal pain, so we performed ileocecal resection as a first treatment. Although it was considered successful, we also performed eradication therapy for *H. pylori* to reduce the possibility of recurrence, because extra gastric MALT lymphomas are suggested to have higher risk of relapse [20].

Conclusion

Colonic MALT Lymphoma is rare, and the optimal therapeutic strategy has not been established. Based on our experience and on previous reports, we suggest it may be feasible to perform surgical resection for patients with mass-forming colonic MALT lymphoma localized without dissemination.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Consent was obtained from the patient for the publication of this report and the accompanying images.

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