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ABOUT COVER

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CASE REPORT

Primary rectal mucosa-associated lymphoid tissue lymphoma treated with only endoscopic submucosal dissection: A case report

Wan-Sik Lee, Myung-Giun Noh, Young-Eun Joo

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Abstract

BACKGROUND

Mucosa-associated lymphoid tissue (MALT) lymphoma is a distinct subtype of non-Hodgkin B cell lymphoma that mostly involves the gastrointestinal tract. The stomach is the most commonly affected site whereas colorectal involvement occurs very rarely. Given its rarity, the management and clinical outcome of colorectal MALT lymphoma are not well established yet.

CASE SUMMARY

From the superficial capillary bed in the lower rectum. Endoscopic ultrasonography showed homogenous hypoechoic lesions in the deep mucosal layer. Endoscopic submucosal dissection (ESD) was done for accurate histologic diagnosis and treatment and both the rectal lesions were completely removed *en bloc* and subsequently diagnosed as primary rectal MALT lymphoma. Herein, we report a case of primary rectal MALT lymphoma in a 68-year-old woman that was treated by only ESD, and the 12-month follow-up revealed no tumour recurrence.

CONCLUSION

These results of our case and previous reports suggest that endoscopic resection alone may be a feasible and safe treatment for primary colorectal MALT lymphoma and allows organ preservation.

Key Words: Rectum; Mucosa-associated lymphoid tissue lymphoma; Endoscopic submucosal dissection

Core Tip: Colorectal involvement of Mucosa-associated lymphoid tissue (MALT) lymphoma occurs rarely and the management of colorectal MALT lymphoma are not well established yet. We report a rare case of colorectal MALT lymphoma treated with endoscopic resection alone. To date, only six cases of primary colorectal MALT lymphomas treated with endoscopic resection alone, including our patient, have been documented in the medical literature. Among the reported cases, there was no recurrence during follow-up. These results of our case and previous reports suggest that endoscopic resection alone may be a feasible and safe treatment for primary colorectal MALT lymphoma and allows organ preservation.

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INTRODUCTION

Gastrointestinal lymphoma is an uncommon disease that constitutes a small proportion of gastrointestinal neoplasms. Primary gastrointestinal mucosa-associated lymphoid tissue (MALT) lymphoma is a rare type of non-Hodgkin lymphoma that comprises 1%-4% of gastrointestinal non-Hodgkin lymphomas[1-3]. Most primary gastrointestinal MALT lymphomas occur in the stomach, and colorectal involvement occurs very rarely. Thus, the management and clinical outcome of colorectal MALT lymphoma are highly variable and not well established[4-7].

Here, we report a case of a 68-year-old woman with primary rectal MALT lymphoma that was treated with endoscopic submucosal dissection (ESD) alone and present a literature review pertaining to this condition.

CASE PRESENTATION

Chief complaints

A 68-year-old woman visited our clinic for a routine health screening examination.

History of present illness

She had no systemic B symptoms, including abdominal pain, fever, night sweats, and weight loss.

History of past illness

Ten years earlier, she underwent surgery for thyroid cancer. She had been on medication for hypertension and diabetes mellitus for 15 years.

Personal and family history

The patient denied any family history of malignant tumours.

Physical examination

On physical examination, she was afebrile, her blood pressure and pulse were normal, and her abdomen was soft, nondistended, and nontender without hepatosplenomegaly or palpable lymphadenopathy.

Laboratory examinations

Laboratory examinations, including complete blood cell count, liver function test, renal function study, and tumour markers, were within normal limits.

Imaging examinations

Computed tomography scan of the neck, chest, abdomen, and pelvis as well as bone marrow aspiration revealed no significant abnormalities.

FURTHER DIAGNOSTIC WORK-UP

Esophagogastroduodenoscopy showed atrophic gastritis with intestinal metaplasia, and the *Campylobacter*-like organism test was negative for *Helicobacter pylori* (*H. pylori*) infection and the patient

didn't have history of previous *H. pylori* eradication therapy. Colonoscopy showed two subepithelial tumours, measuring 4 and 5 mm and arising from the superficial capillary bed into the lower rectum (Figure 1), that seemed to be neuroendocrine tumours. Endoscopic ultrasonography revealed two homogenous hypoechoic lesions in the deep mucosal layer (Figure 2). As rectal neuroendocrine tumor was suspected according to the endoscopic ultrasonography, the two rectal lesions were removed *en bloc via* ESD for accurate histological diagnosis and treatment (Figure 3).

FINAL DIAGNOSIS

On routine histology, haematoxylin–eosin staining showed a dense aggregate of lymphoid cell in the lamina propria layer forming polypoid-lesion (Figure 4A). These lymphoid cells that had small-to-intermediate nuclei and focally clear cytoplasm, infiltrated into muscularis mucosae but did not infiltrate into submucosa (Figure 4B). Immunohistochemistry to ascertain the nature of tumour cells showed positive staining for CD20 (Figure 4C), but negative results for CD3 and Bcl-6. The Ki-67 Labelling index was 5%. Characteristic lymphocyte-epithelial infiltration of CD20-positive tumor cells was also observed (Figure 4D). The biopsy specimens indicated a diagnosis of MALT lymphoma. In accordance with the Ann Arbor staging system, the tumour was diagnosed as a stage IE primary rectal MALT lymphoma.

TREATMENT

The two rectal lesions were removed *en bloc via* ESD.

OUTCOME AND FOLLOW-UP

The patient has been followed-up regularly at the outpatient clinic. Although follow-up period of the patient has been only 12 mo, there was no evidence of recurrence at 12 mo after the ESD.

DISCUSSION

MALT lymphoma is classified as an extranodal marginal zone B-cell lymphoma of the MALT type[1-3] that frequently involves the gastrointestinal tract, including stomach and small bowel, and very rarely involves the colorectal structures[4-7]. Therefore, the clinical characteristics, treatment, and outcome of primary colorectal MALT lymphoma have not been clearly established yet.

The median age at diagnosis of colorectal MALT lymphoma is approximately 60 years, with a slight female predisposition, and the clinical presentation is most often asymptomatic, followed by abdominal discomfort/pain, positive result on a stool occult blood test, constipation, diarrhoea, tenesmus, and obstruction. The most common lesion site is the rectum, followed by the terminal ileum, cecum, and sigmoid colon. The main endoscopic appearance is of a subepithelial tumour, followed by polyposis, ileitis, and epithelial mass type[7]. Our patient is a 68-year-old woman with an asymptomatic rectal MALT lymphoma that comprised two subepithelial tumours that were found incidentally on screening colonoscopy.

Gastric MALT lymphoma is strongly associated with *H. pylori* infection, and *H. pylori* eradication is the main therapeutic strategy in primary gastric MALT lymphoma[1-3]. However, the association between colorectal MALT lymphoma and *H. pylori* infection is unclear. In our patient, the rectal MALT lymphoma was not associated with an *H. pylori* infection.

Colorectal MALT lymphomas were treated by various modalities, including single or a combination of endoscopic resection, surgery, *H. pylori* eradication with antibiotics, radiation therapy, or chemotherapy. The overall prognosis of colonic MALT lymphoma showed an indolent nature and favourable clinical behaviour[4-7]. However, because of its rarity and indolent nature, the treatment and outcome of colorectal MALT lymphoma is not well established.

In our case, ESD was undertaken for accurate histological diagnosis and treatment. Rectal lesions were completely resected *en bloc* by ESD and were pathologically confirmed as a rectal MALT lymphoma. Given the stage IE status of lesions limited to only the rectum, based on a discussion with our multidisciplinary medical team, observation without additional treatment was planned. Twelve months after the ESD, the patient had no tumour recurrence.

The first case of endoscopic resection with a hot-snare guillotine technique was reported in primary rectal MALT lymphoma in 2009, wherein empirical *H. pylori* eradication therapy was added despite a negative result on the *H. pylori* test[8]. Another case of stage IE primary rectal MALT lymphoma was

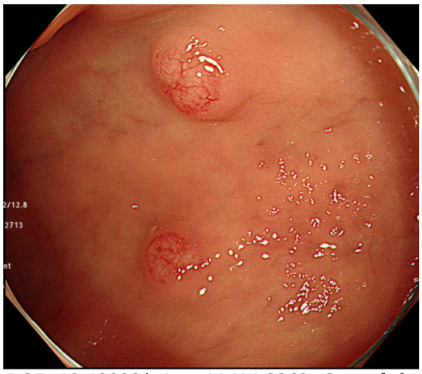

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Figure 1 Colonoscopy shows two subepithelial tumors, measuring 4 and 5 mm, within the superficial capillary bed and rising into the rectum.

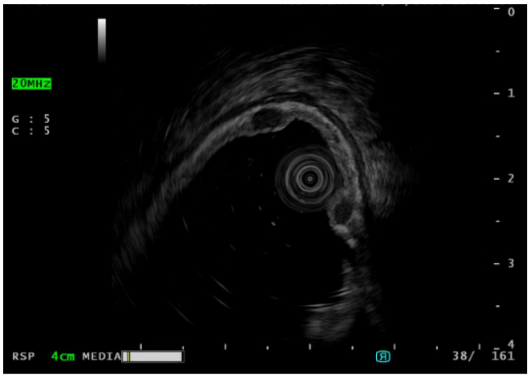

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Figure 2 Endoscopic ultrasonogram shows two homogenous hypoechoic lesions in the deep mucosal layer.

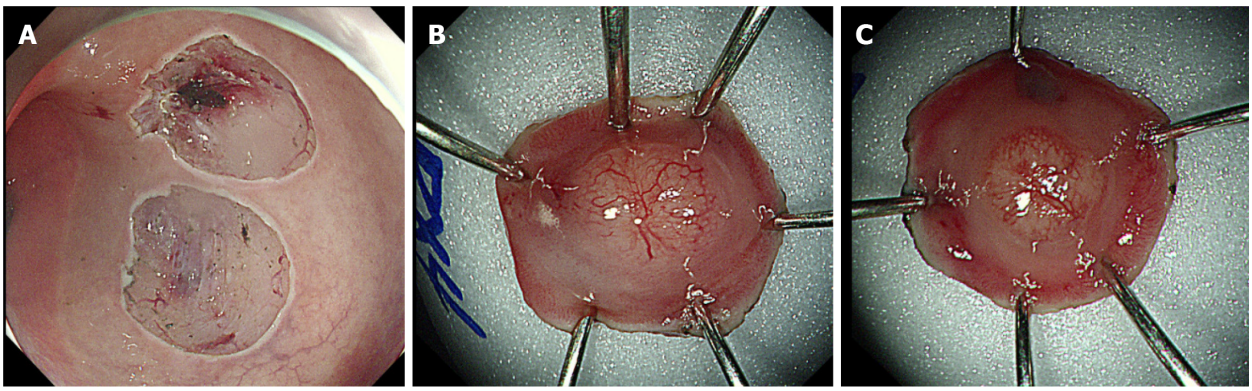

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Figure 3 Two rectal lesions have been removed *en bloc* by endoscopic submucosal dissection. A: Two ESD induced ulcers; B: Tumor size was measured 5 mm × 4 mm and specimen size was measured 12 mm × 12 mm; C: Tumor size was measured 4 mm × 4 mm and specimen size was measured 12 mm × 10 mm.

diagnosed by endoscopic mucosal resection (EMR) and treated with radiation therapy[9].

To date, only six cases of primary colorectal MALT lymphomas treated with endoscopic resection alone, including our patient, have been documented in the medical literature (Table 1)[10-14]. The patients were aged 46 to 72 years (mean age, 64.2 years) and included three men and three women. On clinical presentation, three cases were asymptomatic, two had bleeding, and one had weight loss. Two cases are treated with EMR, two with ESD, one with EMR with ligation, and one with endoscopic full-thickness resection. The mean follow-up period was 30.2 mo (range, 12–60 mo). Among the reported cases, there was no recurrence during follow-up.

Table 1 Summary of reported cases of primary colorectal mucosa-associated lymphoid tissue lymphomas treated with endoscopic resection alone

Patient No.	Ref.	Age (yr)/Sex	Symptoms (duration)	Location	Size (cm)	Endoscopic findings	Treatment	Follow up period (mo)	Outcome
1	Lin *et al* [10], 2016	59/M	Positive fecal occult blood test	Colon (25 cm from anal verge)	2.0	Polypoid lesion with wide base, slightly irregular border, and an irregular vascular pattern with mild inflammatory changes	EMR	36	No recurrence
2	Shah *et al* [11], 2021	72/M	Asymptomatic	Mid rectum	2.0	Raised erythematous lesion	EMR	60	No recurrence
3	Yoon *et al*[12], 2021	69/F	Weight loss (3 mo)	Lower rectum	1.0	Subepithelial tumor	EMR with ligation	37	No recurrence
4	Tao *et al* [13],2022	46/M	Asymptomatic	Rectum (10 cm from anal verge)	3.5	Laterally spreading tumor-like lesion	ESD	24	No recurrence
5	Li *et al* [14], 2022	71/F	Hematochezia (1 mo)	Lower rectum	6.0	Hemispheric mass with rough and hyperemic mucosa	Endoscopic full-thickness resection	12	No recurrence
6	Present case	68/F	Asymptomatic	Lower rectum	0.5, 0.3	Two subepithelial tumors with superficial capillary bed	ESD	12	No recurrence

MALT: Mucosa-associated lymphoid tissue; EMR: Endoscopic mucosal resection; ESD: Endoscopic submucosal dissection.

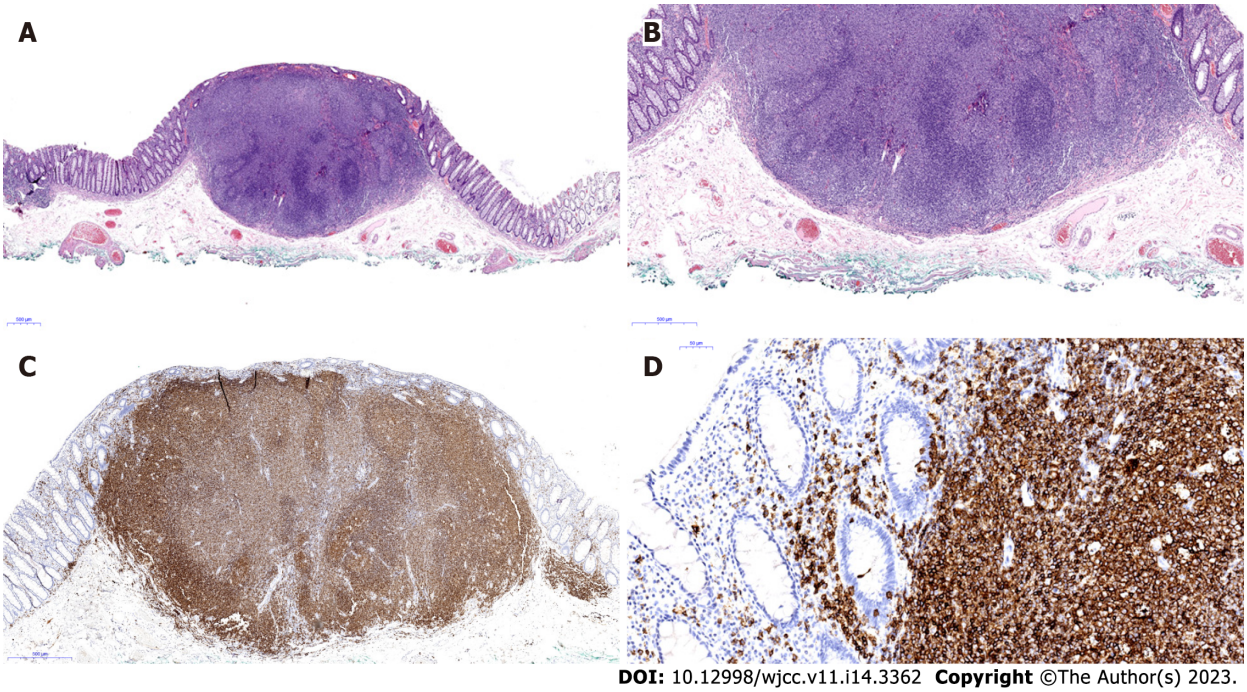

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Figure 4 Microscopic findings. A: Endoscopic biopsy specimens show a dense aggregate of lymphoma cells in the lamina propria (hematoxylin and eosin staining, ×20); B: Lymphoma cells infiltrate the mucosal layer above the subepithelial layer (hematoxylin and eosin staining, ×40); C: Immunohistochemical staining shows aggregate of lymphoma cells that stained positive for the B-cell marker CD20 (×400); D: Characteristic lymphocyte-epithelial lesion of CD20-positive lymphoma cells was also observed (x400).

CONCLUSION

These results of our case and previous reports suggest that endoscopic resection alone may be a feasible and safe treatment for primary colorectal MALT lymphoma and allows organ preservation. However, long-term follow-up data are needed to determine the efficacy of this treatment approach in a larger

number of cases that have been treated with endoscopic resection alone.

FOOTNOTES

Author contributions: Lee WS, Noh MG, Joo YE contributed to manuscript writing and editing and data collection; Joo YE contributed to conceptualization and supervision; all authors have read and approved the final manuscript.

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