

Case Report

Endoscopic mucosal resection for esophageal mucosa-associated lymphoid tissue lymphoma: a case report

Dacheng Wu*, Bin Deng*, Weiming Xiao, Lei Wang, Yanbing Ding

Department of Gastroenterology, The First People's Hospital of Yangzhou, Yangzhou 225000, China. *Equal contributors.

Received January 24, 2016; Accepted April 29, 2016; Epub June 15, 2016; Published June 30, 2016

Abstract: Esophageal mucosa-associated lymphoid tissue (MALT) lymphoma is a very rare disease, and the clinical characteristics have not been clearly defined. Endoscopic mucosal resection (EMR) has been introduced for the treatment of a variety of gastrointestinal diseases, but its therapeutic potential for MALT lymphoma remains uncertain. Here we report a case of primary esophageal MALT lymphoma. A 48-year-old man was referred to our hospital for the evaluation of an esophageal submucosal tumor (SMT) that had been detected incidentally by endoscopy. Endoscopy detected the presence of a longitudinally round and tubular shaped smooth elevated lesion in esophagus. Endoscopic ultrasonography (EUS) also confirmed the discovery of a longitudinally growing hypoechoic mass located in the submucosal layer of the esophagus. Subsequently, he underwent EMR to remove the lesion, and the pathology of the resected specimen diagnosed the mass as primary esophageal B-cell lymphoma of MALT type. In addition, this case report may help further the current understanding on this disease.

Keywords: Mucosa-associated lymphoid tissue, lymphoma, esophagus, endoscopic mucosal resection, diagnosis, therapy

Introduction

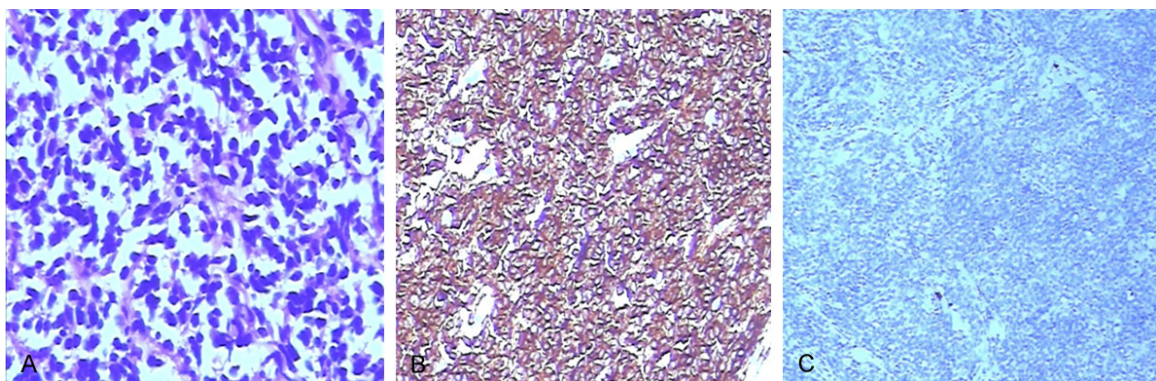
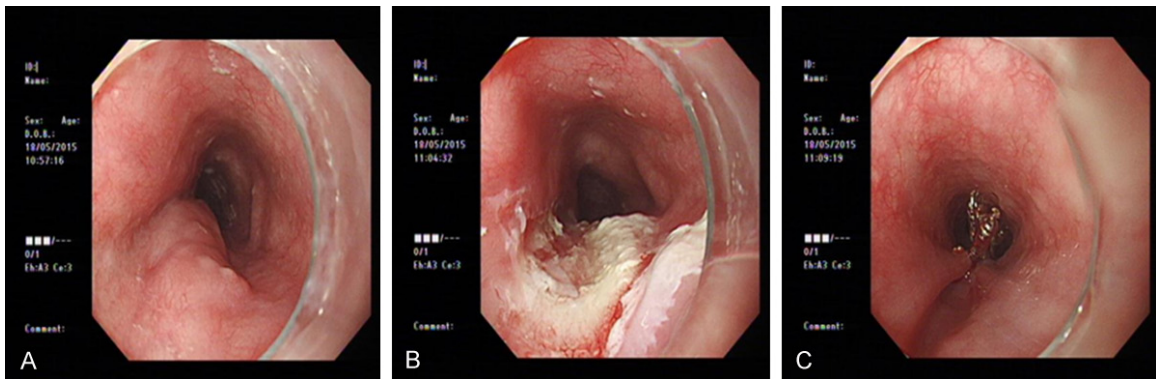
Mucosa-associated lymphoid tissue (MALT) lymphoma is a subtype of B-cell non-Hodgkin lymphoma that was first described by Isaacson and Wright [1]. The most common site of MALT lymphoma is the gastrointestinal tract, but it is also has been found in the lung, breast, bladder, kidney, liver, skin, conjunctiva, thyroid gland, and thymus [2]. Primary esophageal MALT lymphoma has been rarely reported, with less than 1% of MALT lymphomas occurring at this site [2]. In this case report, we describe the treatment of a patient diagnosed with esophageal MALT lymphoma by endoscopic mucosal resection (EMR).

Case presentation

Regular gastroduodenoscopic examination of a 48-year-old male patients showed that the esophageal mucosa was elevated to a distance of 22-24 cm from the incisorteeth (**Figure 1A**).

The fast urease test result was positive, indicating the presence of *Helicobacter pylori* (Hp) infection. The findings of endoscopic ultrasound (EUS) were consistent with those of gastroscopy, and the existence of an elevated esophageal lesion arising from the submucosal layer supported a possible diagnosis of leiomyoma (**Figure 1B**). The patient was admitted to our hospital on May 16, 2015. No abnormalities were detected on physical examination, and chest and abdominal computed topography (CT) scans showed thickening of the wall of the middle esophagus (**Figure 1C**). Therefore, an initial diagnosis of an esophageal submucosal lesion was made, and the patient agreed to undergo esophageal EMR (**Figure 2**). Proton pump inhibitors were administrated after the procedure. Pathological examination confirmed the diagnosis of esophageal MALT lymphoma with muscularis mucosae infiltration, and the borders of the resected specimen were free of tumor cells (**Figure 3A**). Immunohistochemical staining showed that the resected tissue

Figure 1 consists of three panels. Panel A is an endoscopic image showing a large, polypoid lesion in the sigmoid colon. Panel B is an OCT image of the lesion, showing a cross-section with a color scale from 0 to 20 cm. Panel C is a CT scan of the colon, showing a large polypoid lesion in the sigmoid colon.



was CD20(+), CD117(-), and dog-1(-) (**Figure 3B, 3C**). No abnormalities were identified upon bone marrow cytology and chromosome karyotyping. A bone marrow biopsy was also conducted and showed that granulocytic proliferation was decreased and juvenile cells were present. Considering all of these results together,

er, the patient was finally diagnosed with esophageal MALT lymphoma. The patient then took a combination of rabeprazole, amoxicillin, clarithromycin, and bismuth potassium citrate to eradicate the Hp infection. At 1-month and 3-month follow-ups, no tumor relapse was detected by gastroduodenoscopy (**Figure 4**).

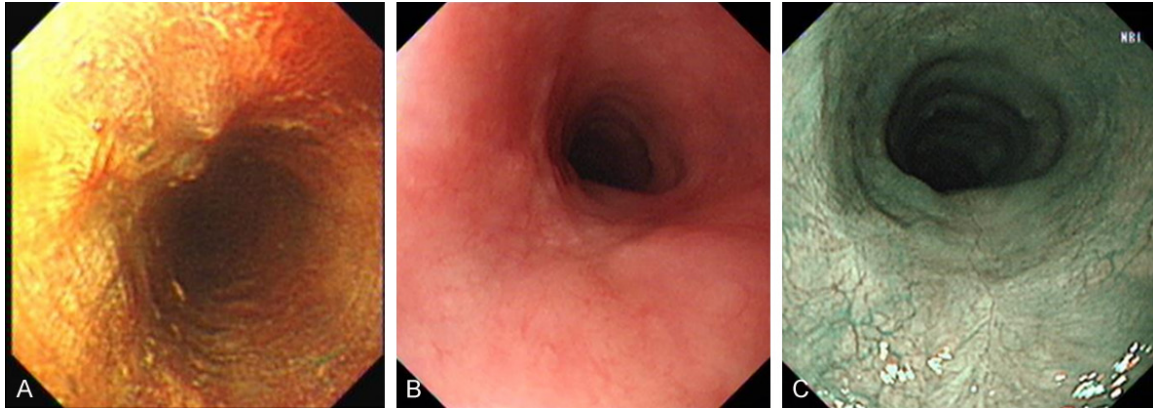


Figure 4. Gastroendoscopic examination showed no sign of relapse or recurrence at 1-month (A) and 3-month (B, C) follow-ups.

Discussion

The clinical features and therapeutic options for esophageal MALT lymphoma have not been fully investigated due to its very low incidence. It is hypothesized that esophageal MALT lymphoma is associated with mechanical stimuli (e.g., food, hot water), chemicals, drugs, chronic inflammation (e.g., gastro-esophageal reflux, Barrett's esophagus), autoimmune disorders (e.g., Hashimoto thyroiditis, lymphocytic interstitial pneumonia) [3, 4]. Patients with esophageal MALT lymphoma usually present with unspecific symptoms, and some patients complain of dysphagia. Gastroendoscopy and EUS are considered the routine diagnostic methods in clinical practice. In such patients, a polyp, an ulcer, mucosal thickening, and stenosis could be observed by gastroendoscopy [5-7]. The lesion may be hyperechoic or hypoechoic in the mucosal or submucosal layer under EUS. Kudo et al. proved that narrow band imaging can be used to diagnose esophageal MALT lymphoma, which is characterized by dendritic vessels in the background of whitish mucosa [3]. In addition, esophageal MALT lymphoma is highly suspected if the lesion grows longitudinally with a whitish surface or as a flat elevated area, resembling keratosis and acanthosis, and if immunohistochemical analysis reveals the tissue to be CD20(+), CD10(+), Bcl2(+), Bcl6(-), CD3(-), CD5(-), and cyclin D1(-). In the present case, a spindle-like submucosal elevation was identified during gastroendoscopy, and EUS findings were consistent with a hypoechoic submucosal lesion. Immunohistochemical staining showed that the cells of the lesion were CD20(+), CD117(-), and dog-1(-).

Based on all of these results together, the patient was finally diagnosed with esophageal MALT lymphoma.

Treatment options for esophageal MALT lymphoma include surgery, radiotherapy, chemotherapy, and endoscopic interventions. EMR and endoscopic submucosal dissection (ESD) may be administered for treatment of early-stage MALT lymphoma, but chemotherapy is the preferred modality for advanced lesions. Generally, MALT lymphomas are sensitive to radiotherapy at a dosage of 20-40 Gy, and most patients tolerate this treatment well [8]. Bardisi et al. treated one patient with esophageal MALT lymphoma by open surgery, and no recurrence or relapse was observed over 12 months of follow-up [9]. Overall, there is still no consensus regarding the management of MALT lymphoma. In the current case, no infiltration was observed in the bone marrow, supporting the diagnosis of early-stage esophageal MALT lymphoma. Thus, EMR was performed to remove the lesion.

It is also known that Hp infection can contribute to the development of MALT lymphoma. However, whether Hp infection can induce esophageal MALT lymphoma is unclear. Kudo et al. reported four patients with esophageal MALT lymphoma, and all were negative for Hp infection [3]. However, because the fast urease test result was positive in our patient, it remains possible that Hp infection might be associated with esophageal MALT lymphoma. Thus, anti-Hp eradication medication was administered.

Here we report a case of esophageal MALT lymphoma, an uncommon disease without specific

Case of esophageal mucosa-associated lymphoid tissue lymphoma

symptoms that must be differentiated from other esophageal submucosal tumors in clinical practice. Our results suggest that endoscopic resection can be an effective therapeutic option for esophageal MALT lymphoma, especially for early stage disease.

Disclosure of conflict of interest

None.

Address correspondence to: Yanbing Ding, Department of Gastroenterology, The First People's Hospital of Yangzhou, Yangzhou 225000, China. Tel: 86-514-82981199; E-mail: chinadbin@126.com

References

- [1] Isaacson P and Wright DH. Malignant lymphoma of mucosa-associated lymphoid tissue. A distinctive type of B-cell lymphoma. *Cancer* 1983; 52: 1410-1416.
- [2] Thieblemont C, Berger F and Coiffier B. Mucosa-associated lymphoid tissue lymphomas. *Curr Opin Oncol* 1995; 7: 415-420.
- [3] Kudo K, Ota M, Narumiya K, Shirai Y, Ohki T and Yamamoto M. Primary esophageal mucosa-associated lymphoid tissue lymphoma treated by endoscopic submucosal dissection. *Dig Endosc* 2014; 26: 478-481.
- [4] Hosaka S, Nakamura N, Akamatsu T, Fujisawa T, Ogiwara Y, Kiyosawa K, Hidaka E, Ota H, Katsuyama T and Inagaki H. A case of primary low grade mucosa associated lymphoid tissue (MALT) lymphoma of the oesophagus. *Gut* 2002; 51: 281-284.
- [5] Herrmann R, Panahon AM, Barcos MP, Walsh D and Stutzman L. Gastrointestinal involvement in non-Hodgkin's lymphoma. *Cancer* 1980; 46: 215-222.
- [6] Papaxoinis G, Papageorgiou S, Rontogianni D, Kaloutsi V, Fountzilas G, Pavlidis N, Dimopoulos M, Tsatalas C, Xiros N and Economopoulos T. Primary gastrointestinal non-Hodgkin's lymphoma: a clinicopathologic study of 128 cases in Greece. A Hellenic Cooperative Oncology Group study (HeCOG). *Leuk Lymphoma* 2006; 47: 2140-2146.
- [7] Taal BG, Van Heerde P and Somers R. Isolated primary oesophageal involvement by lymphoma: a rare cause of dysphagia: two case histories and a review of other published data. *Gut* 1993; 34: 994-998.
- [8] Sugimoto M, Kajimura M, Shirai N, Furuta T, Kanaoka S, Ikuma M, Sato Y and Hishida A. Outcome of radiotherapy for gastric mucosa-associated lymphoid tissue lymphoma refractory to *Helicobacter pylori* eradication therapy. *Intern Med* 2006; 45: 405-409.
- [9] Bardisi ES, Alghanmi N and Merdad AA. Primary mucosa-associated lymphoid tissue lymphoma of the esophagus masquerading as a benign tumor. *Ann Med Surg (Lond)* 2014; 3: 39-42.