

Gastrointestinal involvement by mantle cell lymphoma identified by biopsy performed during endoscopy

A case report

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Abstract

Rationale: Primary gastrointestinal mantle cell lymphoma is rare, and histopathological examination and specific immunohistochemical staining are still the gold standard for diagnosis. Therefore, it is necessary to find a new way to improve positive biopsy rates.

Patient concerns: A 58-year-old man was admitted to our hospital with epigastric pain, abdominal distension, nausea, and melena. Endoscopy identified submucosal neoplasms and diffuse gastrointestinal tract involvement including the esophagus.

Diagnoses: A false-negative diagnosis was first determined by ordinary endoscopy. However, a large tissue biopsy was subsequently performed using endoscopic mucosal resection based on endoscopic ultrasonography (EUS). Pathological examination of the biopsy specimens taken from the lesions of the duodenum and rectum revealed diffuse lymphocytic proliferation and obscure nodular and small cleaved cells with irregularly shaped nuclei. Immunohistochemistry showed that the cells were positive for CyclinD1, BCL-2, CD20, CD21, and CD5; however, they were negative for CD3, CD6, CD10, and CD43.

Interventions: The patient refused to receive further treatment.

Outcomes: Mantle cell lymphoma was conclusively diagnosed.

Conclusions: EUS has an important role in the diagnosis and management of gastrointestinal submucosal tumors. Performing a pathological biopsy including EUS may be useful for identifying the unknown nature of tumors of the digestive tract.

Abbreviations: EMR = endoscopic mucosal resection, EUS = endoscopic ultrasonography, MCL = mantle cell lymphoma.

Keywords: endoscopic mucosal resection, endoscopy, gastrointestinal, mantle cell lymphoma

1. Introduction

Mantle cell lymphoma (MCL) is an aggressive B-cell lymphoma derived from a subset of naive prenominal center cells that has a propensity for involving extranodal sites. Primary gastrointestinal MCL is rare. We report the case of a 58-year-old man who was admitted to our hospital with epigastric pain, abdominal distension, nausea, and melena. He was ultimately diagnosed with MCL. This case suggests that endoscopic ultrasonography

(EUS) and pathological biopsy may be useful for identifying tumors of an unknown nature in the digestive tract. This case will hopefully help others gain more knowledge and, in turn, experience regarding the endoscopic diagnosis of MCL originating from the gastrointestinal tract.

2. Methods

We obtained the patient's medical records and reviewed the related literature. Informed consent to participate in the study was obtained from the patient. This study was approved by the People's Hospital of Guangxi Zhuang Autonomous Institutional Review Board.

3. Clinical summary

A 58-year-old man was admitted to our hospital for epigastric pain, abdominal distension, nausea, and melena. Physical examination was unremarkable and revealed no palpable mass or lymphadenopathy. Laboratory findings revealed anemia (hemoglobin, 6.4 g/dL) with an increased erythrocyte sedimentation rate (140 mm/h) and C-reactive protein (73.83 mg/L) level. Biochemical findings obtained by performing liver function tests, renal tests, and tumor markers (CEA, CA 199) were all within normal limits. Endoscopy identified numerous submucosal lesions in the esophagus, whole stomach, and duodenum, along with some erosion in the gastric antrum. Colonoscopy showed

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Figure 1. Endoscopy identified multiple polypoid tumors in the whole stomach (A), duodenum (B), colon (C), and rectum (D).

multiple polypoid tumors (5–20 mm) in the intestinal segments, including the rectum and terminal ileum, and some had hyperemia (Fig. 1). Pathological examination of the biopsy specimens from the gastric antrum showed chronic inflammation with ulcer, intestinal metaplasia, and lymphatic follicle formation (Fig. 2). EUS (Olympus EUS EU-ME1 Miniprobe sonography) demonstrated that the lesions were almost homogeneously hypoechoic neoplasms originating from the submucosal layer, with most of them breaking through the mucosal muscle layer. The bulk biopsy specimens from the duodenum and rectum underwent endoscopic mucosal resection (EMR), and the wounds were sealed with titanium clips (Fig. 3). Pathological examination of the biopsy specimens showed diffuse lymphocytic proliferation and obscure nodular and small cleaved cells with irregularly shaped nuclei. The cells were positive for CyclinD1, BCL-2, CD20, CD21, and CD5; however, they were negative for CD3, CD6, CD10, and CD43. Ki-67 staining revealed a proliferative index of 20% (Fig. 4). Based on these results, a diagnosis of Ann Arbor stage IV MCL was confirmed. However, the patient refused to receive further treatment.

4. Discussion

MCL most commonly affects men and usually presents in the fifth or sixth decade of life.^[1] Primary gastrointestinal MCL is rare, accounting for only 1% to 4% of primary gastrointestinal lymphoma.^[2] There are no specific clinical manifestations, but the most frequent symptoms are abdominal pain, diarrhea, palpable masses, melena, hematochezia, weight loss, and fatigue.^[3,4] Any part of the gastrointestinal tract may be involved

in MCL, from the stomach to the colorectum, and it can be diffuse or restricted to the intestinal tract with isolated lesions.^[5,6] Our case showed diffuse gastrointestinal tract involvement including the esophagus. Using ordinary endoscopy, MCL appears in the forms of mucosa ulcers, polypoid type changes, masses, and others.^[7] It is mainly characterized by homogeneously hypoechoic lesions that originate from the submucosa but do not affect the propria on EUS.^[8] As the disease progresses, its original hierarchy will gradually disappear.^[9] Histopathological

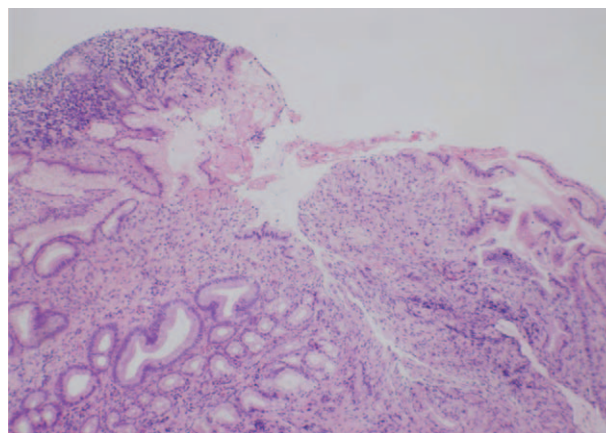


Figure 2. The first diagnosis was false-negative. Pathological examination of the biopsy specimens from the gastric antrum showed chronic inflammation with ulcer, intestinal metaplasia, and lymphatic follicle formation.

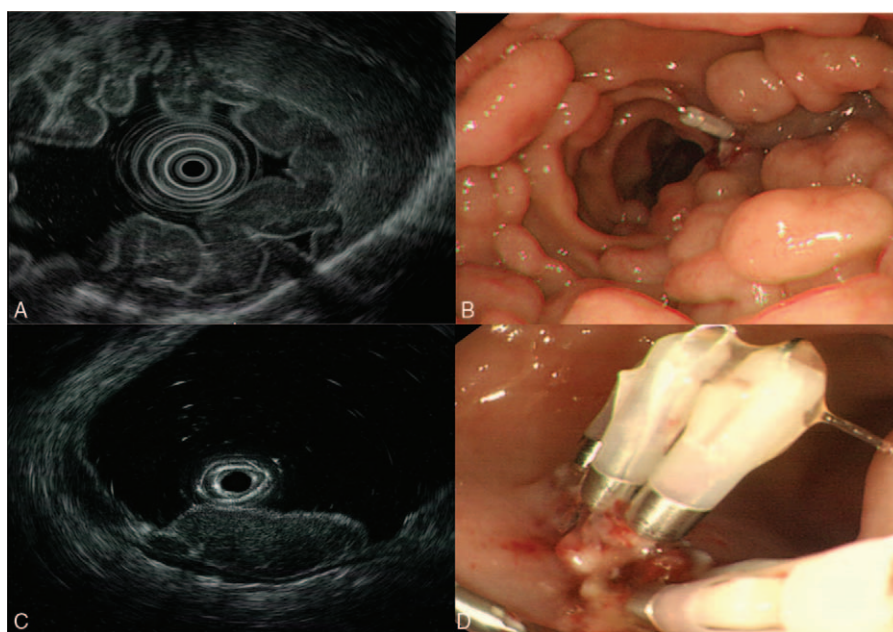


Figure 3. Endoscopic ultrasonography demonstrated that the lesions were almost homogeneously hypoechoic neoplasms originating from the submucosal layer (A and C). Bulk biopsy specimens taken from the duodenum (B) and rectum (D) underwent EMR. EMR = endoscopic mucosal resection.

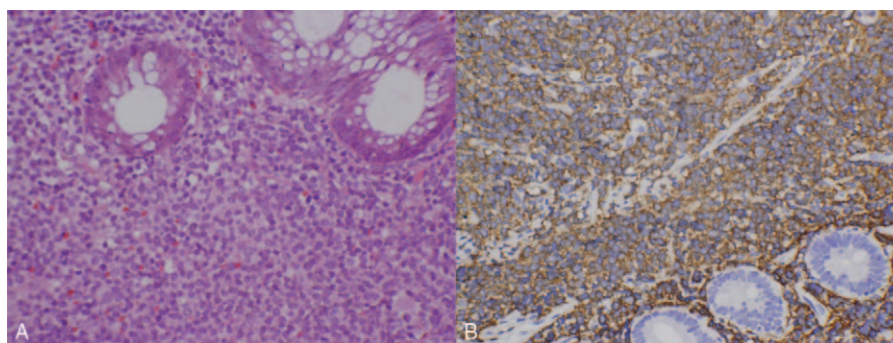


Figure 4. Pathological examination revealed diffuse lymphocytic proliferation and obscure nodular and small cleaved cells with irregularly shaped nuclei (A). Immunohistochemistry showed that cells were positive for CyclinD1, BCL-2, CD20, CD21, and CD5 (B).

examination and specific immunohistochemical staining are still the gold standard for diagnosis. However, because the lesions originate from deep within the mucosa or submucosa, mucosal biopsy using common endoscopy may not be able to detect the lesions. In fact, our patient received a false-negative diagnosis when ordinary endoscopy was used during the first examination. A subsequent large tissue biopsy was performed using EMR based on EUS and was able to define the tumor origin. The patient received a definitive diagnosis after histopathological examination of the biopsy specimens revealed atypical lymphoid proliferation consistent with MCL lymphoma. Therefore, performing a biopsy using EUS purposefully may improve the diagnostic rate.

MCL is an aggressive, incurable type of non-Hodgkin lymphoma. Many cases are advanced when they are confirmed. The complete remission rate of chemotherapy is low, and the median duration of remission is 1.5 to 3 years.^[10] Because MCL

is associated with a poor prognosis, early diagnosis is emphasized. EUS is a minimally invasive method with an important role in the diagnosis and management of gastrointestinal submucosal tumors. MCL presenting as submucosal lesions on EUS has been rarely reported. Performing EUS and a pathological biopsy may be useful for identifying the unknown nature of tumors of the digestive tract. Further studies using these methods are required to obtain more information.

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