CASE REPORT

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Treatment of Solitary Extramedullary Plasmacytoma of the Stomach with Endoscopic Submucosal Dissection

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Extramedullary plasmacytoma (EMP) is an uncommon manifestation of plasma cell neplasia, which occurs outside the bone marrow. Intestinal involvement usually occur secondarily in multiple myeloma but it occur primarily less commonly. We experienced a woman who had a primary EMP localized in the stomach and the EMP was removed successfully by endoscopic submucosal dissection. The lesion was pathologically confirmed as confined to the gastric mucosa. **(Gut and Liver 2009;3:334-337)**

Key Words: Endoscopic submucosal dissection; Extramedullary; Plasmacytoma

INTRODUCTION

Extramedullary plasmacytoma (EMP) is uncommon and represents about 3% of all plasma cell neoplasms. Almost 90% of EMP occurs in the head and neck area in the upper respiratory tract; the gastrointestinal tract is a very rare site of involvement of this neoplasm.[1-3] Because EMP is highly sensitive to radiotherapy, surgery of head and neck disease is not routinely recommended. However, when it occurs as a non-upper respiratory disease, surgery is not inferior to radiotherapy.[3,4] Endoscopic submucosal dissection (ESD) is one type of endoscopic resection; it is a minimally invasive treatment modality for early-stage gastrointestinal neoplasms and is currently accepted in many countries as standard treatment for early gastric carcinoma (EGC) confined to the mucosa.[5,6] ESD is less invasive and more economical than surgery, and allows complete histopathological staging.[5,7,8] Here we present the first case of a gastric solitary EMP limited to the mucosa treated by ESD.

CASE REPORT

A 50-year-old woman was admitted to our hospital because of abnormal findings on an esophagogastroduodenoscopy performed for routine medical evaluation. The endoscopy showed well-demarcated, flat, yellow-whitish mucosal changes on the anterior and posterior walls of the lower body of the stomach about 3 cm and 1 cm in diameter, respectively (Fig. 1). The microscopic findings of the endocopic biopsies revealed extensive infiltration of plasma cells with intranuclear immunoglobulin inclusion that were consistent with the diagnosis of plasmacytoma (Fig. 2). The complete blood count and routine biochemistry were normal. After admission, bone marrow aspiration/biopsy and peripheral blood smear were performed and there was no evidence of clonal marrow or peripheral plasmacytosis. Skull X-rays, abdomen and pelvic computed tomography (CT) scans showed no underlying bone lesions or lymph node enlargement. Serum and urine immunoelectrophoresis demonstrated normal density of albumin, the $\alpha 2$ fraction and β fraction, and the serum and urine immunofixation showed no specific monoclonal bands. The patient underwent endoscopic ultrasonography (EUS) to evaluate the depth of the plasmacytoma. There was abnormal mucosal thickening but no evidence of tumor invasion into the submucosa (Fig. 3). Therefore, we diagnosed the patient with a solitary EMP of the stomach and an ESD was performed (Fig. 4).

En bloc resection was carried out without complications such as bleeding or perforation. The ESD specimens

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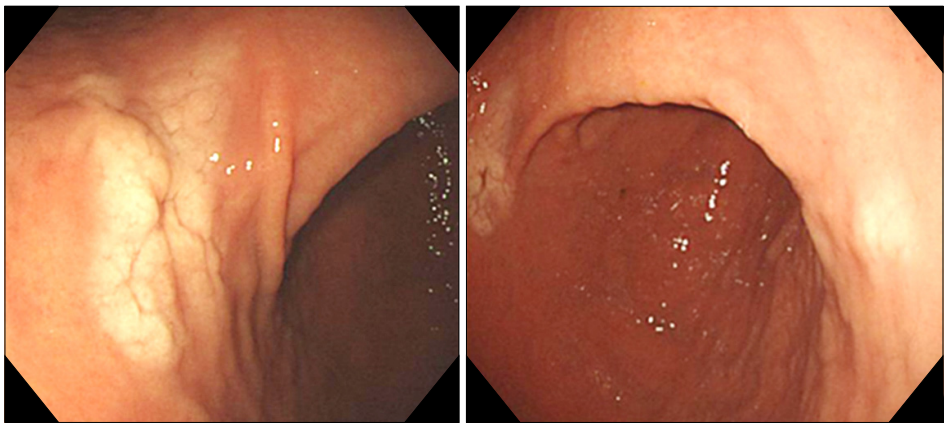

Fig. 1. Esophagogastroduodenoscopy. Well-demarcated, flat, yellow-whitish mucosal changes on the anterior and posterior walls of the lower body of the stomach.

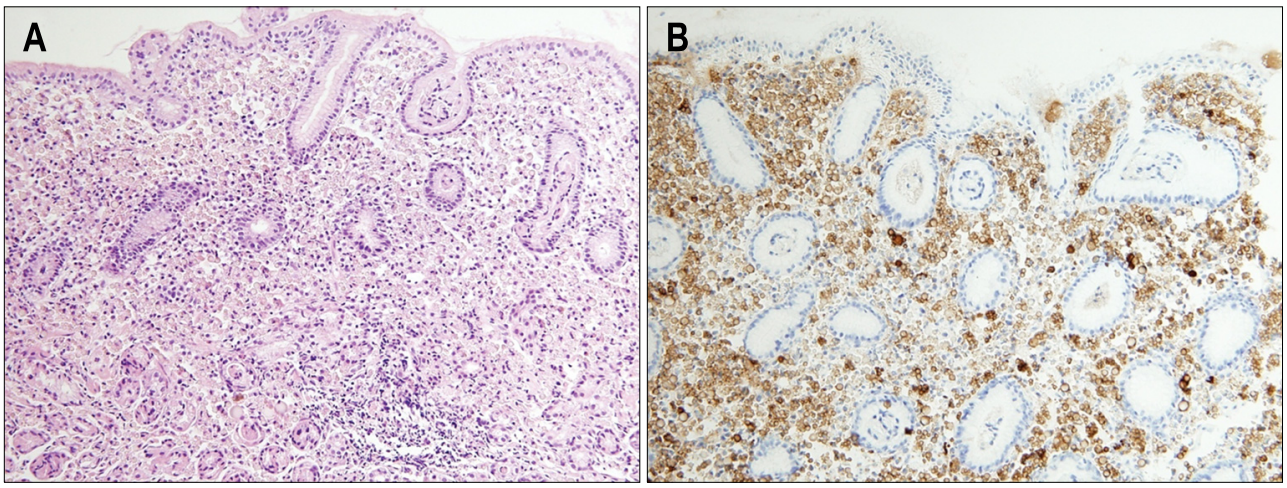

Fig. 2. (A) The constituent tumor cells consist of mature plasma cells together with plasmablast, which have large, central, prominent nucleoli. (B) Immunohistochemistry showing positivity for lamda chains confirming monoclonal nature of tumor.

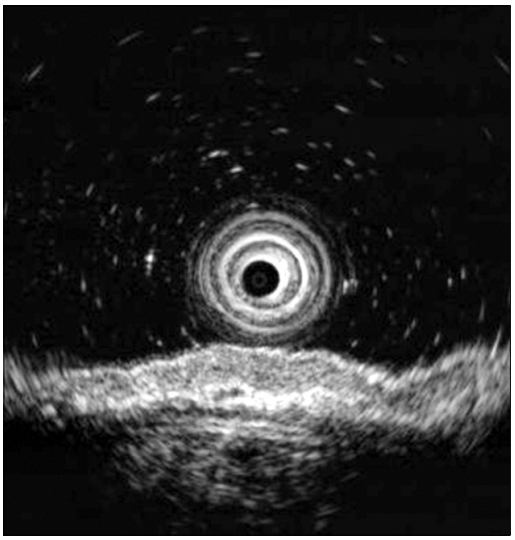

Fig. 3. Endoscopic ultrasonography finding showed extramudullary plasmacytoma of stomach confined to the mucosa.

showed plasma cell infiltration and abundant intranuclear immunoglobulin inclusion with positive immunohistochemistry for lamda chains that was limited to the mucosa with clear vertical margins (Fig. 5). Three months after the ESD, a follow up endoscopy was performed. Changes associated with scaring were present at the ESD site. Multiple biopsies of the ESD scar sites and surrounding tissue were performed. The microscopy showed normal gastric mucosa and no plasma cell infiltration of the scars and the surrounding tissue. Endoscopic biopsy, abdomen and pelvic CT, routine laboratory findings, and the peripheral blood smear 6 and 12 months after treatment also demonstrated no evidence of tumor recurrence or development of systemic myeloma.

DISCUSSION

Solitary EMP is less common than solitary plasmacytoma of the bone (SBP), and accounts for approximately

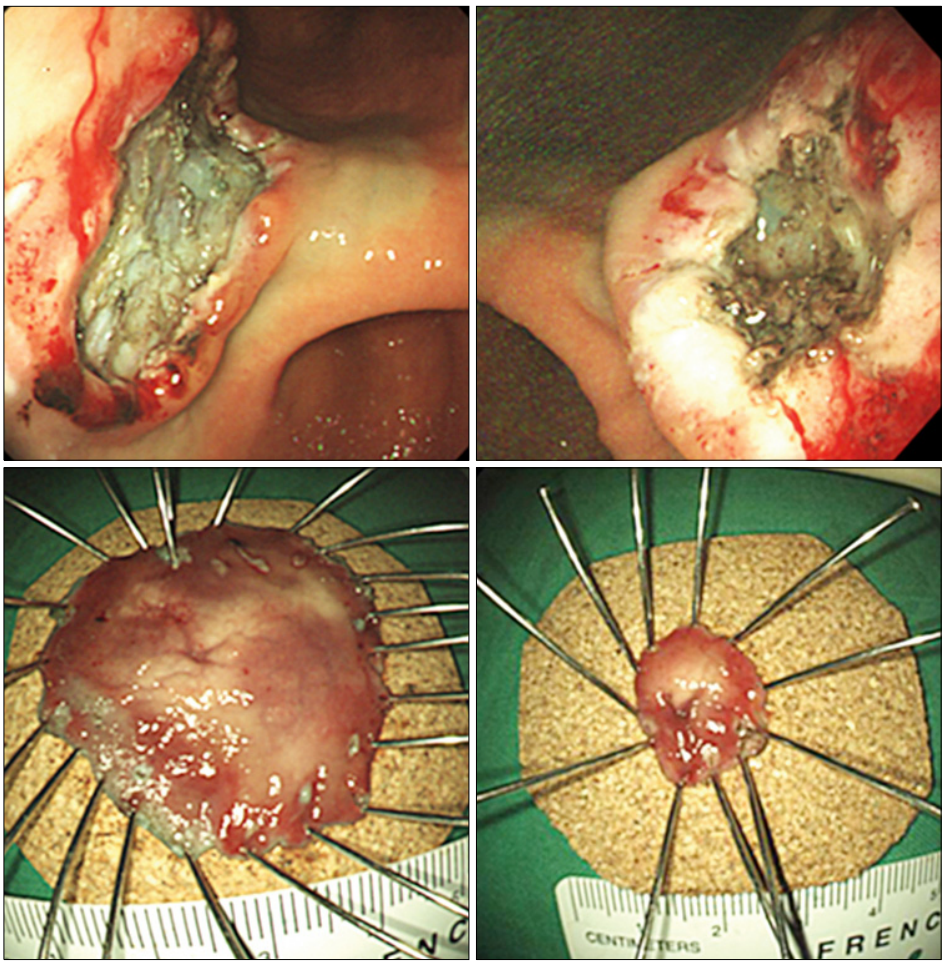

Fig. 4. Endoscopic submucosal dissection was performed with no significant complication.

3% of all plasma cell neoplasms.[1,2] Almost 90% of cases with EMP develop in the head and neck area, especially in the upper respiratory tract including the sinuses, nasal cavity, nasopharynx, and tonsils. The gastrointestinal tract is a very rare site for involvement of this neoplasm.[1-3] The diagnosis of EMP is based on morphology and immunohistochemical demonstration of monoclonal κ or λ light chains or heavy chains found in localized plasma cell tumors in the absence of plasma cell proliferation elsewhere, especially in the bone marrow, and must be distinguished from reactive plasmacytomas, plasma cell granulomas and lymphomas.[2-4] After curative treatment, about 70% of patients with solitary EMP remain disease-free at 10 years, and fewer than 30% of patients with EMP develop systemic progression.[1-4]

Currently, there are no general guidelines for the treatment of patients with EMP. However, EMP is highly sensitive to radiotherapy, and surgery is not recommended for the initial treatment of head and neck disease because of its invasiveness.[3,4] However, for disease at other sites, including the gastrointestinal tract, surgery reveals no significant difference in survival outcomes compared to radiotherapy.[3] There have been some cases reported with a solitary gastrointestinal plasmacytoma that was treated by surgery, bortezomib and dexamethasone, laparoscopic surgery, or *Helicobactor pylori* eradication.[9-11] However currently, endoscopic mucosal resection has been accepted as a reliable treatment for EGC limited to mucosa. ESD, a new method of endoscopic resection, was developed in order to increase en bloc resection rate, for larger lesions. But there still exists a limitation of ESD for infiltrative disease. This is because the safety margin is just a few millimeters in most cases. In our case the lesion was relatively well demarcated to perform ESD. We performed ESD to treat solitary gastric EMP confined to the mucosa that was documented by EUS; this is the first case reported. We observed no significant side effects associated with the ESD and no documented relapse over 12 months after treatment. ESD has the advantages of being less invasive, less expensive, and has a lower incidence of side effects compared with conventional surgery or radiotherapy.[5,6,12]

Further study is needed to evaluate the outcomes of treatment of solitary gastric EMPs by ESD. Our findings

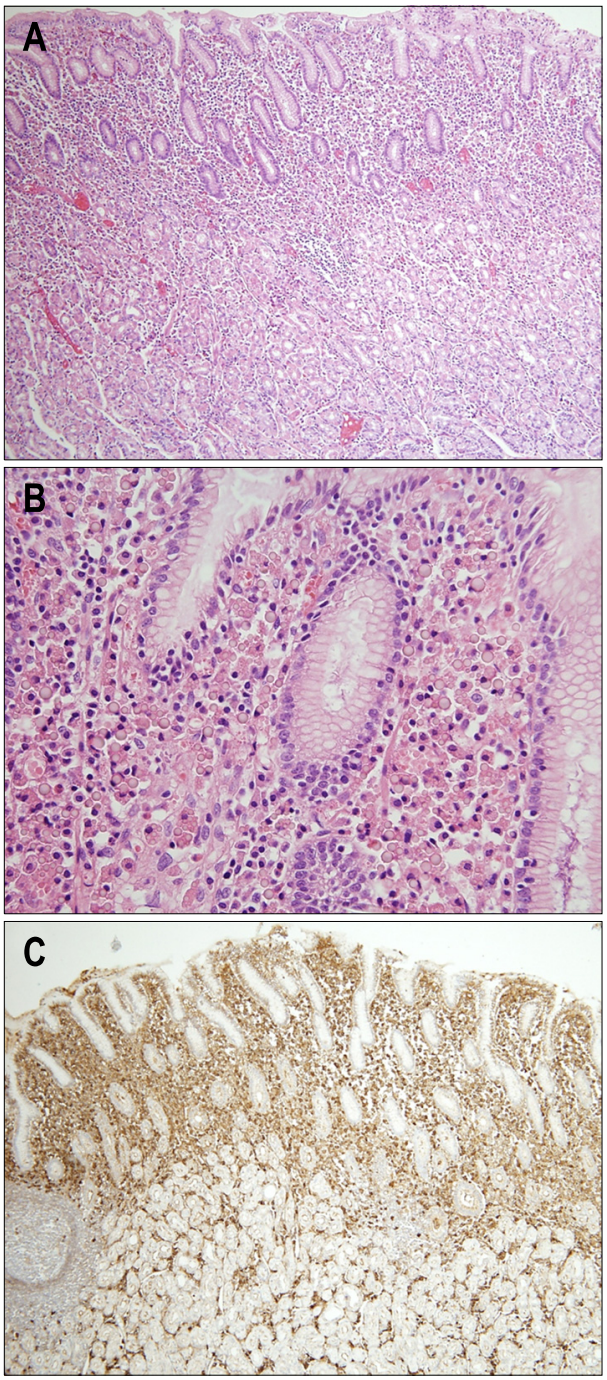

Fig. 5. (A) Low-magnification view of an endoscopic submucosal dissection specimen indicating the presence of abundant plasma cells confined to mucosa (H&E stain, ×100). (B) High-magnification view of an endoscopic submucosal dissection specimen indicating intranuclear immunoglobulin inclusion (H&E stain, ×400). (C) Immunohistochemistry showing positivity for lamda chains confirming monoclonal nature of tumor.

suggest that ESD is safe, well-tolerated and an effective alternative treatment for solitary gastric EMP confined to the mucosa.

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