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LETTER TO THE EDITOR

Endoscopic submucosal dissection combined with orally administered dimethyl sulfoxide for primary gastric localized amyloidosis

Dear editor,

The deposition of amyloid within the gastrointestinal tract is a common characteristic of systemic amyloidosis [1]; however, primary gastric amyloidosis is extremely rare [2]. The prognosis for patients diagnosed with primary gastric localized amyloidosis (PGLA) remains poor, despite currently available non-specific therapeutic approaches, including administration of dimethyl sulfoxide (DMSO), nutritional support, and surgery. Unfortunately, none of these approaches yields significant benefits in clinical practice.

A 33-year-old female patient presented with complaints of epigastric pain that had commenced 3 weeks prior. Initially, she developed epigastric discomfort and dyspepsia with heartburn and acid reflux, which was associated with occasional epigastric pain. Within a week of the onset of discomfort, the patient's symptoms worsened. A physical examination and laboratory tests revealed no abnormalities. Endoscopic examination revealed a lesion with irregular borders and mucosa in the lesser curvature of the gastric body, measuring approximately 1.2×1.2 cm (Fig. 1A_{1-2}). The lesion was associated with superficial, diffuse, small patches of hemorrhagic patches in the mucosa of the gastric body and fundus, and an additional lesion with regular borders and a smooth mucosal surface in the gastric fundus measuring 1.0×2.0 cm (Fig. 1B_{1-2}). Hematoxylin and eosin staining revealed a structureless deposition of amyloid that extended from the submucosal layer to the muscularis propria, and large amounts of amyloid were also found around the small blood vessels of both lesions. The amyloid deposition was negative for van Gieson staining and positive for Congo red staining (Fig. 1). Multiple biopsies from the esophagus, duodenum, colon, and bone marrow demonstrated no amyloid deposition. Subsequently, the patient underwent an endoscopy ultrasound evaluation with a 7.5-MHz echoendoscope (UMP230; Olympus Optical Co. Ltd., Tokyo, Japan). A normal gastric wall was observed in five ultrasonographic layers. The lesion in the mucosa of the lesser curvature of the gastric body had a hypoechoic stratum mucosum and lamina muscularis mucosae with increased thickness extruding into the gastric cavity. The local muscularis propria was intact. The second lesion in the gastric fundus was non-echogenic and was traceable through the submucosa of stratum mucosum (Fig. 2). Computed tomography imaging revealed a parenchymal goiter with a diffuse spindle shape on the posterior wall of the gastric body that measured 4.0×1.1 cm and was accompanied by a low-density cystic area (25 Hu; Fig. 2).

A diagnosis of PGLA was made. An endoscopic submucosal dissection procedure was performed using a hook knife (KD-260LR; Olympus Optical Co. Ltd., Japan) and an IT knife (KD-610L; Olympus Optical Co. Ltd., Japan) with a high-frequency generator (ICC200; Erbe, Tubingen, Germany) through a standard single-accessory-channel endoscope (GIF-H260; Olympus Optical Co. Ltd., Japan), as described previously [3]. The margin of the lesion was defined by an argon-air knife. Marking dots were placed on the normal mucosa approximately 5 mm away from the lesion, and direct views of the submucosal layer were possible. Injections of saline and epinephrine were inserted into the submucosal layer around the lesion margin to elevate the mucosa [4]. A mucosal incision was then made outside the marking dots with a hook knife. Direct dissection of the submucosal layer beneath the lesion was performed under direct vision to obtain tissue samples using IT knife. During the endoscopic submucosal dissection (ESD) operation under general anesthesia, the exposed blood vessels of the lesion beds were coagulated thoroughly with diathermy forceps (Fig. 3). The lesions were harvested using aspiration and evaluated histologically. The aspirated lesions were similar to the pre-treatment biopsies and the diagnosis of PGLA was confirmed. After ESD, the patient was designated nil by mouth and received intravenous fluids and omeprazole (40 mg bid).

Two weeks postoperatively, a repeat endoscopy revealed ulceration of the gastric mucosa (Fig. 4). DMSO was administered orally for 6 months after the ESD procedure (33.3% solution in water or fruit juice, batch number Q/HG314499; Tianjin Bodi Chemical Industry Co. Ltd., China) in accordance with previous reports [5]. The initial dose was 3.0 g/d, which was gradually increased up to the maximum tolerable dose of 20.0 g/d. The total dose was 3.0—20.0 g/d, and administered in three equal doses after meals. Follow-up

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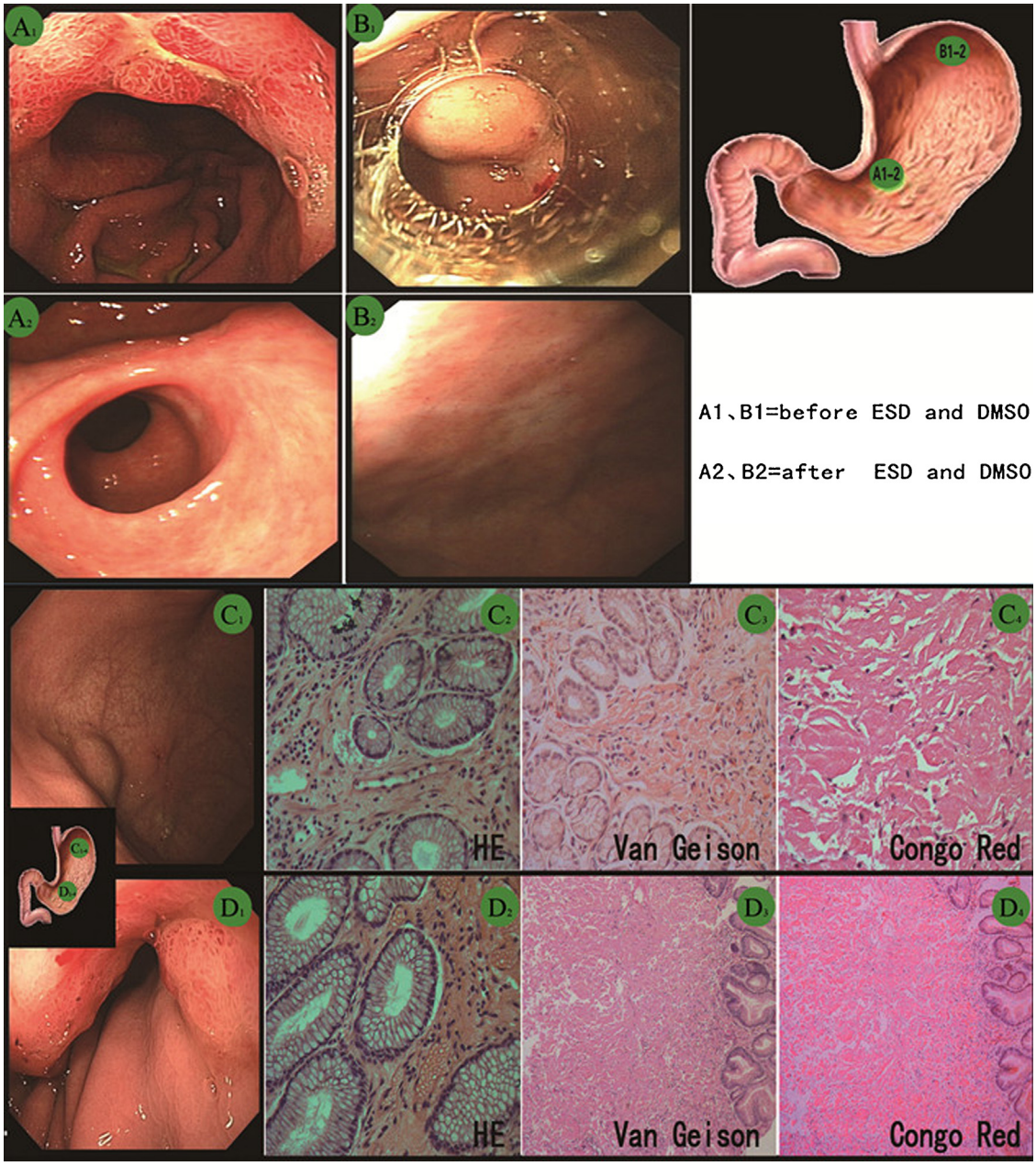

Figure 1 Endoscopic findings pre- and post-treatment and histological examination of biopsied gastric body and gastric fundus. A 1.2×1.2 cm irregular protuberance with an eroded irregular surface and borders was observed in the lesser curvature of the gastric body. Pathological samples were taken from here (A_1). A 1.0×1.0 cm submucosal mass was found in the gastric fundus with a smooth surface in an area of good gastric mobility. The endoscope with a monocyclic COOK ligator was inserted into the stomach and targeted to the lesion. The surface mucosa of the tumor was dissected by the ligator, but the tumor was too large to be extracted and the endoscope was removed (B_1). Scars with smooth surfaces and normal surrounding mucosa were observed at the gastric fundus (A_2), lesser curvature of the gastric antrum and angle of the stomach (B_2) postoperatively. Active bleeding and fistulae were not observed. The surface of the surrounding mucosa was smooth and its appearance was normal. An irregular protuberance with an eroded irregular surface and borders was observed in the gastric body (C_1). The section showed that there was a glass-like deposition in the deep lamina propria of the gastric mucosa and submucosa, and some surrounding the small blood vessels (C_2). The deposition stained positive for Congo red (C_3) and van Gieson (C_4) staining, and the medullary thyroid carcinoma tissue was used as a control. Compared to the control group, the deposition was considered to be a starch-based substance rather than collagen fibers.