A Pedunculated Submucosal Lesion in the Stomach with Inverted Downgrowth

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Abstract

A 70-year-old man with a gastric lesion was referred to our hospital because of an unusual pedunculated lesion in the gastric body. Endoscopic ultrasound showed scattered cystic lesions within a heterogeneous area confined to the submucosal layer. Endoscopic mucosal resection was performed to obtain a precise diagnosis, as well as for removal. The lesion was histopathologically diagnosed as a heterotopic submucosal gland. We herein describe this rare type of gastric polyp and provide a literature review.

Key words: gastric submucosal lesion, histopathology, endoscopic mucosal resection

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Introduction

A heterotopic gastric gland is a relatively rare lesion characterized by the ectopic proliferation of gastric glandular elements, typically beneath a normal mucosal layer. This lesion is commonly misdiagnosed because of its histopathological characteristics (1). This lesion has previously been described as a hamartomatous polyp, hamartoma mimicking a submucosal tumor (SMT), hamartomatous inverted polyp and heterotopic inverted polyp (2-5). A similar lesion has recently been called an inverted hyperplastic polyp (IHP) because of its similarity to its colonic counterpart (6), and more than 30 cases of gastric IHP have been reported (7, 8). Although these submucosal lesions have been considered to be benign in nature, the pathogenesis of these lesions is not fully understood, and histopathological criteria have not been established. Moreover, several gastric IHPs accompanied by adenocarcinoma have recently been reported (2, 7-9). In this report, we describe a gastric submucosal lesion with inverted downgrowth which was diagnosed and treated by endoscopic mucosal resection (EMR).

Case Report

A 70-year-old man with a gastric lesion was referred to our hospital after gastric cancer screening. Gastroscopy identified a pedunculated lesion, approximately 10 mm in diameter, as well as multiple fundic gland polyps in the gastric body (Fig. 1a). No atrophic gastritis was noted. The lesion seemed to be covered with normal gastric mucosa with no inflammatory or erosive changes when it was turned using forceps (Fig. 1b). Bridging of surrounding folds was not evident. The patient was suspected to have a SMT. Endoscopic ultrasound (EUS) was performed using a catheter probe (20 MHz), and it showed scattered cystic lesions within a heterogeneous area confined to the submucosal layer (Fig. 2). The lesion was considered to be an aberrant pancreas, lymphangioma, inflammatory fibroid polyp or IHP, but it was difficult to make a definitive diagnosis based on these findings.

EMR was performed in order to make a precise diagnosis, as well as for removal of the tumor. The tumor was completely removed en bloc without any adverse events. Histopathologically, the lesion consisted of hypertrophic smooth muscle bundles with glandular structures and cystic forma-
Gastroscopy identified a pedunculated lesion of 10 mm in size in the gastric body (a). The lesion seemed to be covered with normal gastric mucosa, with no inflammatory or erosive changes (b).

EUS showed scattered cystic lesions within a heterogeneous area confined to the superficial submucosal layer.

The lesion consisted of hypertrophic smooth muscle bundles with glandular structures and cystic formation (Hematoxylin and Eosin staining).

These pyloric glands showed irregular branching and dilatation (Hematoxylin and Eosin staining).

demonstrated strong positivity for MUC6 (gastric pyloric gland type secretory mucin), and weak positivity for MUC5AC (gastric surface type secretory mucin).

Discussion

This solitary pedunculated polyp with a cystic gastric gland demonstrated several unique characteristics, including hypertrophic smooth muscle bundles with marked branching surrounding the pyloric glandular structures, glands showing irregular branching, dilatation and cystic formation. No dysplastic area was present. A heterotopic submucosal gland, which is the term used in the Japanese Classification of Gastric Carcinoma 14th edition, seemed to be a suitable description for this pathological entity (9).

A similar lesion was recently called an IHP in other reports (2, 7, 8). Although several gastric IHPs accompanied by adenocarcinoma have been reported recently, histopathological criteria have not been established for these submucosal lesions, including hamartomatous polyps, hamarto-
IHP, because it resembles IHP of the colon.

To date, five cases of gastric IHP with coexisting adenocarcinoma have been reported (2, 7, 8, 10), although the pathogenesis remains unclear. Yamashita et al. (2) suggested that IHP may be gastritis cystica profunda (GCP) associated with cystic dilatation of the glandular epithelium, leading to a polypoid lesion. The main lesion is located in the submucosa or the inside of the muscularis mucosae. In 1993, Kamata et al. (6) named the lesion IHP, because it resembles IHP of the colon.

The clinical and endoscopic characteristics of the reported cases of gastric IHPs accompanied with adenocarcinoma are shown in Table. There was one case of IHP accompanying adenocarcinoma without GCP or Helicobacter pylori infection. We decided to remove the present lesion, because of IHP accompanied with adenocarcinoma could not be eliminated based only on the endoscopic and endosonographic appearance, although there was no GCP or Helicobacter pylori infection.

With regard to the morphology of gastric IHP, there are two types: IHP without a stalk, which are the SMT type, and IHP with a stalk, which is the polytype (5). Polytpe IHP can be removed by endoscopic resection (4, 8), while surgical resection should be considered for the SMT type lesions or larger lesion of any type, because of the difficulty of removing it in one piece (5, 7). Considering the possibility of coexisting adenocarcinoma in gastric IHP, en bloc resection is preferred to avoid residual cancer cells and relapse (11). In our case, EUS showed scattered cystic lesions within a heterogeneous area that was 10 mm in size confined to the superficial submucosal layer. EMR was considered to be safe and efficient for removing it in one piece.

It has been suggested that p53 dysregulation may play an important role in the malignant transformation of IHP (7). However, defining the process of malignant transformation within a gastric IHP presents a challenge, because the lesion is extremely rare. When an unusual subepithelial lesion is found and gastric IHP is suspected, such a unique lesion should be considered for removal, including removal (preferably in one piece) by EMR and ESD, to allow a precise diagnosis to be made. Over-treatment should be avoided, but a histopathological assessment is indispensable unless coexisting adenocarcinoma has been ruled out.

In conclusion, we herein described a rare case of a gastric submucosal lesion presenting inverted downgrowth, and have provided a review of the literature about similar lesions. To clarify the pathogenesis and establish classifications according to the gastric submucosal heterotopic gland, further investigations will be required. Gastroenterologists should be more aware of such unique lesions in the stomach.

The authors state that they have no Conflict of Interest (COI).

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References


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**Table. Characteristics of Gastric Inverted Hyperplastic Polyp Accompanied with Adenocarcinoma**

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age/Sex</th>
<th>Site</th>
<th>Gross</th>
<th>Size (mm)</th>
<th>EUS finding</th>
<th>GCP</th>
<th>HP infection</th>
</tr>
</thead>
<tbody>
<tr>
<td>2</td>
<td>69/M</td>
<td>Body</td>
<td>Y-I</td>
<td>10 × 9</td>
<td>not shown</td>
<td>+</td>
<td>-</td>
</tr>
<tr>
<td>2</td>
<td>81/M</td>
<td>Formix</td>
<td>Flat elevation</td>
<td>5 × 5</td>
<td>not shown</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>7</td>
<td>54/M</td>
<td>Antrum</td>
<td>Pedunculated</td>
<td>45 × 35</td>
<td>heterogeneous tumor</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>8</td>
<td>40/F</td>
<td>Body</td>
<td>Protrusion</td>
<td>35 × 32 × 18</td>
<td>heterogeneous and hyperechoic</td>
<td>-</td>
<td>unknown</td>
</tr>
<tr>
<td>10</td>
<td>72/M</td>
<td>Body-Cardia</td>
<td>Y-II*</td>
<td>35 × 35</td>
<td>disruption of submucosal layer</td>
<td>+</td>
<td>+</td>
</tr>
</tbody>
</table>

*: Yamada-Fukuoka classification, GCP: gastritis cystica profunda, HP: Helicobacter pylori