

A Rare Case of Cystic Subepithelial Tumor in the Stomach: Gastric Adenomyoma

드문 낭성 위상피하 종양에 대한 증례 보고: 위샘종

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Gastric adenomyoma is a rare benign subepithelial tumor, characteristically composed of mucosal structures and a prominent smooth muscle stroma. Because of rarity and the nonspecific computed tomography (CT) features, it is difficult to diagnose gastric adenomyoma before operation. In our case, gastric adenomyoma showed a well-circumscribed cystic subepithelial mass with uneven wall thickness on a CT scan, similar to the findings of former reports. The radiologic differential diagnosis can be narrowed down to several diseases, including duplication cysts, gastritis cystica profunda, brunner's gland hyperplasia and solid tumors with cystic degeneration. Also, adenomyoma could be included in the differential diagnosis of gastric cystic subepithelial masses, especially in the distal part of the stomach.

Index terms

Gastric Adenomyoma
Cystic Subepithelial Tumor
Computed Tomography
Endoscopy

INTRODUCTION

Gastric adenomyoma is a rare benign subepithelial tumor, characteristically composed of mucosal structures and a prominent smooth muscle stroma (1). The majority of the cases are asymptomatic and are found incidentally during endoscopic examinations, or on computed tomography (CT) scans. However, this disease entity is unfamiliar to endoscopists and radiologists, and it is difficult to diagnose before operation. In this report, we describe a case of a gastric adenomyoma, and present its imaging features.

CASE REPORT

A 28-year-old man with a history of epigastric pain spanning a few days was referred to our hospital for a gastric subepithelial

lesion found on the endoscopic examination. His past medical history was unremarkable. A physical examination did not reveal any significant abnormalities. The routine laboratory tests were within the normal ranges.

An endoscopic examination revealed a round mass protruding into the lumen, covered with relatively normal mucosa, in the gastric antrum (Fig. 1). The mass was smoothly compressible by an endoscopic forceps, but an endoscopic biopsy was not conducted. Subsequently, an abdominal CT scan was performed. The CT scan showed a 3.5 cm sized well-circumscribed cystic mass that protruded into the gastric antrum. The lesion had enhancing walls, with uneven thickness, and an internal content of homogeneous density (Fig. 2). No enlarged lymph nodes around the stomach were observed.

A decision for surgical treatment was made for a histologic diagnosis. A laparoscopic wedge resection for the subepithelial

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mass was performed under general anesthesia.

A macroscopic examination revealed a 38 × 35 × 25 mm nodular mass with a yellowish brown color. It was solid and cystic, with clear, mucinous fluid in the lumen. A microscopic examination showed the hypertrophic smooth muscle fibers of the stroma, surrounded by epithelial elements in the form of individual glandular structures (Fig. 3). A final histologic diagnosis was compatible with gastric adenomyoma. The postoperative progress of the patient was uneventful.

DISCUSSION

Adenomyoma of the gastrointestinal tract is a rare benign tumor composed of glands and cysts, lined by columnar, flattened

epithelia and a prominent smooth muscle stroma (1). They are most frequently found in the stomach and duodenum (2). In the stomach, the majority of cases involve the distal part of the stomach (antrum 85%, pylorus 15%) (3). The age of the patients at presentation ranges from 1 week to 81 years, and two-thirds of the cases occur in the fourth to sixth decades (3, 4).

Because there are several different opinions on the histogenetic origin, there are various terms used for adenomyomas: myoepithelial hamartoma, myoglandular hamartoma, adenomyomatous hamartoma, and adenomyosis. Several authors have proposed that gastric adenomyoma is considered to be a variant of an ectopic pancreas without exocrine or endocrine components, based on the similarities of the epithelial components (1). Takeyama et al. (5) placed more weight on the abnormal smooth

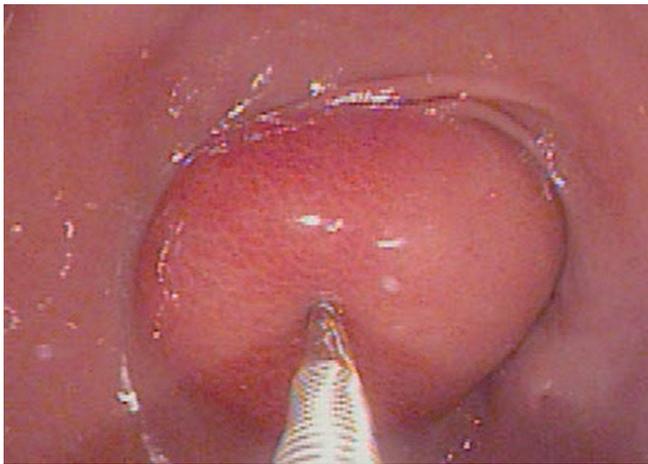


Fig. 1. An upper gastrointestinal endoscopic examination shows a round mass protruding in to the lumen in the anterior wall of the gastric antrum. Relatively normal overlying mucosa is observed, and the mass is smoothly compressible by an endoscopic forceps.

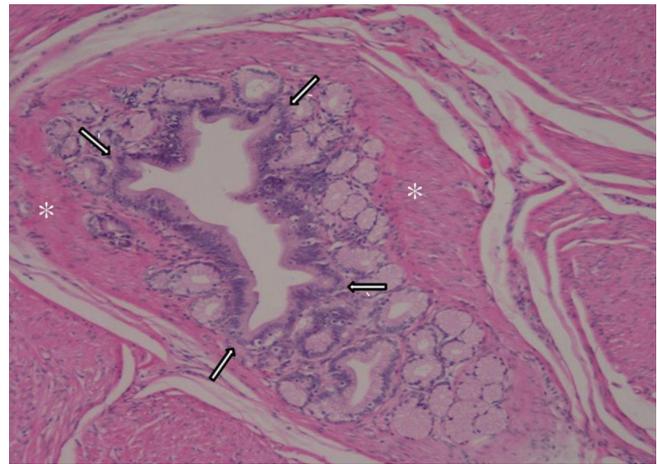
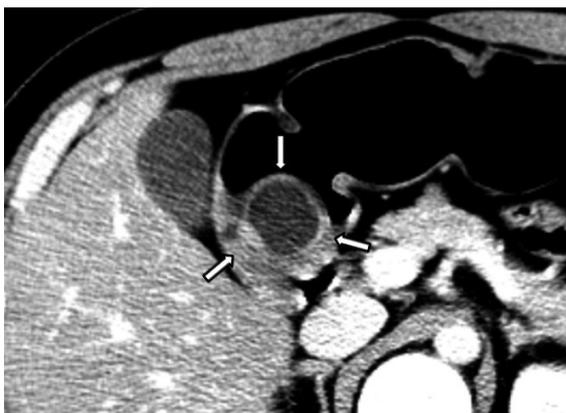


Fig. 3. A microscopy of the specimen shows epithelial elements (arrows) in the form of individual glandular structures supported by the hypertrophic smooth muscle fibers (*) of the stroma (hematoxylin & eosin stain, × 50).



A



B

Fig. 2. Axial (A) and coronal (B) reformatted contrast-enhanced CT images show a well-circumscribed cystic mass (arrows), with uneven wall thickening, in the gastric antrum.

muscle proliferation and considered it to be an hamartoma, resulting from an abnormal interaction between the endoderm- and mesoderm-derived tissues.

Some previously reported cases of gastric adenomyoma demonstrated CT findings, which were identical to the findings in our case, including a well-circumscribed and mainly cystic subepithelial mass that protrudes into the lumen and has irregular or uneven wall thickness (6, 7). The cystic portion of the lesions is correlated with the mucinous component, on the histopathologic examination. Although these findings are nonspecific for subepithelial lesions of the stomach, a differential diagnosis can be narrowed down to several cystic subepithelial lesions: duplication cysts, gastritis cystica profunda and Brunner's gland hyperplasia. Solid subepithelial tumors that can undergo cystic degeneration, such as gastrointestinal stromal tumor and schwannoma can also show similar CT features.

Upon an endoscopic examination, the adenomyoma showed a round bulging mass with overlying normal mucosa that occasionally had central umbilication or ulceration (6). An endoscopic biopsy of the lesion is not useful because the lesions are usually located in the submucosa (1). Recently, endoscopic submucosal dissection has widely been used for subepithelial tumors, and can be the alternative choice for diagnosis and treatment (8).

Although gastric adenomyoma is usually asymptomatic, or has nonspecific symptoms, some complications or symptoms can occur, including localized peritonitis, melena, and gastric outlet obstruction (2, 5, 9). Furthermore, a few cases associated with adenocarcinoma have been reported (3). The relationship between adenomyoma and adenocarcinoma remains unclear. However, the possibility of the malignant transformation of adenomyoma is suggested.

As mentioned above, because of rarity of gastric adenomyoma, non-specific CT and endoscopic findings and uncertain result of endoscopic biopsy, it is difficult to differentiate adenomyoma before operation. Therefore, surgical excision is performed for histologic diagnosis in most cases. During operation, frozen sections are useful for intraoperative diagnosis and avoiding unnecessary radical operation (2). No recurrence after a suc-

cessful surgical resection has been reported.

In summary, an adenomyoma is a rare subepithelial tumor of the stomach that shows nonspecific CT features. It resembles several diseases, including duplication cysts, gastritis cystica profunda, Brunner's gland hyperplasia and solid tumors with cystic degeneration, in radiologic findings. So, adenomyoma could be included in the differential diagnosis of gastric cystic subepithelial masses, especially in the distal part of the stomach.

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드문 낭성 위상피하 종양에 대한 증례 보고: 위샘종

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위샘종은 특징적으로 점막 구조와 풍부한 평활근으로 이루어진 매우 드문 양성 상피하 종양이다. 이 질병의 희귀성과 비특이적인 컴퓨터단층촬영 소견 때문에 수술 전에 위샘종을 진단하기는 어렵다. 저자들의 증례에서 위샘종은 컴퓨터단층촬영에서 이전 보고들과 비슷하게 경계가 잘 지어지는 불규칙한 벽을 가진 낭성 상피하 종양으로 나타났다. 방사선적 감별 진단은 중복낭, 심재성 낭성 위염, 브루너샘 과다형성, 낭성 변성을 동반한 고형 종양으로 좁혀질 수 있다. 유사한 영상소견을 보이는 위샘종도 위의 낭성 상피하 종양에서, 특히 위의 원위부에 있을 경우에 감별진단에 포함되어야 하겠다.

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