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TEACHING CASE

Adenomyoma with goblet and Paneth cells of the ileum

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Abstract

A case of ileal adenomyoma with goblet and Paneth cells is reported. A 75-year-old man died of ruptured hepatocellular carcinoma. As an incidental finding at autopsy, a $9 \times 7 \times 6\,mm^3$-sized nodule was found in the ileal wall. Histologically, the lesion occupied the submucosa and muscularis propria, and consisted of glandular structures of various sizes and interlacing smooth muscle bundles surrounding the glandular elements. Goblet cells and Paneth cells were interspersed in the glandular element. Immunohistochemically, the glandular element was positive for cytokeratin (CK) 7 and negative for CK 20. This is the first reported case of adenomyoma of the gastrointestinal tract that contained Paneth cells. The result of the immunohistochemical staining favored the heterotopic pancreas theory concerning its pathogenesis. The appearance of goblet and Paneth cells might be the result of metaplasia.

Keywords: Adenomyoma; Cytokeratin; Gastrointestinal tract; Goblet cell; Paneth cell

Introduction

Adenomyoma of the gastrointestinal (GI) tract is a rare benign tumor-like lesion that is histologically characterized by a mixture of glandular structures lined by tall columnar epithelium and intervening smooth muscle bundles. It has several synonyms such as myoepithelial hamartoma [4,8,10,18,21,23], adenomyomatous hamartoma [6,13], and foregut choristoma [11]. It usually occurs in the pyloric region of the stomach or duodenum, and its occurrence in the small intestine distal to the duodenum is very rare. A careful review of the literature found only 21 reported cases of adenomyoma in this region [2,4,6–8,10,11,13–19,21–24]. Although goblet cells are occasionally found in the glandular element, there have been no reports on adenomyoma of the GI tract that contained Paneth cells. The pathogenesis of adenomyoma of the GI tract is generally considered to be either a form of hamartoma or a pancreatic heterotopia. In this paper, we report the first case of ileal adenomyoma accompanied by Paneth cells. We discuss the pathogenesis of this lesion based on the results of immunohistochemical staining.

Clinical summary

A 75-year-old man was admitted to our university hospital due to bradycardia. Abdominal computer tomography demonstrated an extensive low-density area of the liver. The serum levels of alpha-fetoprotein (AFP) and protein induced by vitamin K absence or antagonist-II (PIVKA-II) were abnormally high (AFP, 90 000 ng/ml; normal value, <20 ng/ml; PIVKA-II, 133 000 mAU/ml; normal value, <40 mAU/ml). Thus, the patient was diagnosed to have hepatocellular carcinoma. Viral markers, i.e., hepatitis B surface antigen (HBsAg) and hepatitis C virus antibody

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(HCVAb), were both negative. Radical treatment was given up, and the patient died 5 days after admission due to exacerbation of the general condition. Autopsy was perfomed 3 h post-mortem.

Pathological results

Gross appearance

At autopsy, 1300 ml of bloody ascites was observed. An extensive yellowish tumor occupied both lobes of the liver, showing a rupture at S3. Metastatic lesions were found in the lungs and regional and mediastinal lymph nodes. As an incidental finding, a nodule ($9 \times 7 \times 6\,mm^3$) was found in the ileal wall (75 cm proximal to the ileo-cecal valve). The lesion did not cause intussusception. The cause of death was thought to be rupture of the hepatic tumor.

Microscopic appearance

The hepatic tumor was a moderately to poorly differentiated hepatocellular carcinoma showing a thick trabecular growth pattern. Multinucleated giant tumor cells and globular hyaline bodies were scattered. Neither chronic hepatitis nor liver cirrhosis was observed in the non-tumorous liver tissue.

The ileal nodular lesion occupied submucosa and muscularis propria. It consisted of glandular structures of various sizes and interlacing smooth muscle bundles surrounding the glandular elements (Figs. 1 and 2). Some glands were cystically dilated. The glandular structures were lined by tall columnar epithelium with basally oriented nuclei, and goblet and Paneth cells were interspersed (Figs. 3a, b). Glands containing goblet cells were small in number, and they were distributed mainly in the shallow portion of submucosa. The number of glands containing Paneth cells was about 10% of all glandular structures, and they were also distributed mainly in the shallow portion of submucosa (Fig. 4). Those glands were surrounded by interlacing smooth muscle bundles, and they were clearly the component of the tumor-like lesion. Neither nuclear atypia nor desmoplastic stroma was observed. The lesion was not accompanied by ectopic pancreatic acini or islets.

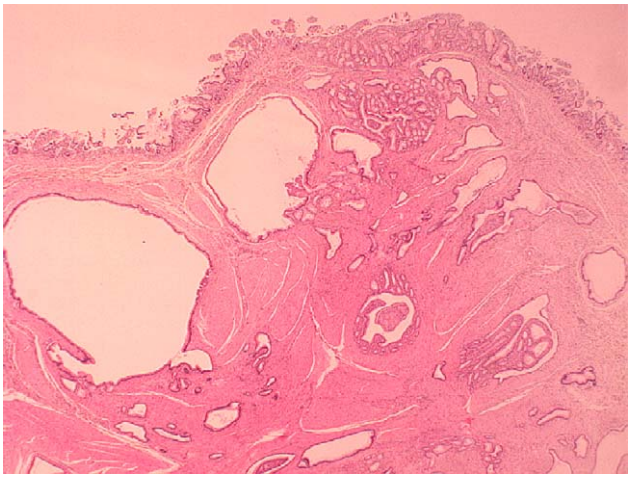

Fig. 1. (Hematoxylin–eosin stain): Low-power view of the ileal adenomyoma. A nodular lesion occupies the submucosa and muscularis propria of the ileal wall. × 20.

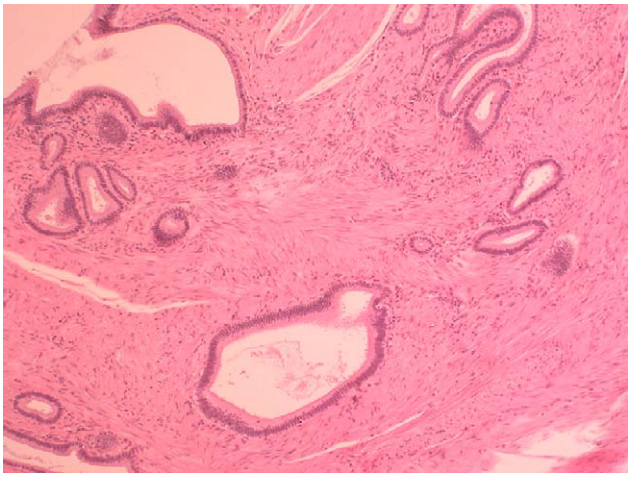

Fig. 2. (Hematoxylin–eosin stain): High-power view of the ileal adenomyoma. The lesion consists of glandular structures of various sizes and interlacing smooth muscle bundles surrounding the glandular elements. × 100.

Immunohistochemical staining was performed on formalin-fixed, paraffin-embedded tissue using the avidin–biotin-peroxidase complex method. The antibodies used were against cytokeratin (CK) 7 (clone OV-TL 12/30, dilution: 1:50; DakoCytomation, Glostrup, Denmark), CK 20 (clone Ks 20.8, dilution: 1:25; DakoCytomation), desmin (clone D33, dilution: 1:50; DakoCytomation), and alpha-smooth muscle actin (clone 1A4, dilution: 1:25; DakoCytomation). Immunohistochemically, the glandular element was positive for CK 7 and negative for CK 20 (Figs. 5a, b). Normal mucosal epithelial cells around the lesion were negative for CK 7 and positive for CK 20. Smooth muscle cells surrounding the glandular elements were diffusely positive for desmin and alpha-smooth muscle actin.

Discussion

The above-mentioned histological and immunohistochemical findings suggested the pathological diagnosis of ileal adenomyoma. Differential diagnoses included enteritis cystica profunda, pneumatosis cystoides