Case Report

Adenomyoma of the Small Intestine in an Adult: Report of a Case

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Abstract

We report a case of adenomyoma in the small intestine, which is an extremely rare entity. An 81-year-old woman presented to our hospital with a history of three episodes of vomiting accompanied by abdominal pain. Upper gastrointestinal examination via a long tube found intestinal obstruction caused by a tumor of the small intestine. Laparotomy revealed a hard mass, 160 cm distal to the Treitz ligament. Pathological examinations of the resected tumor confirmed a diagnosis of adenomyoma originating in the small intestine. To our knowledge, this is only the second report of an adenomyoma of small intestine causing intestinal obstruction in an adult.

Key words Adenomyoma · Small intestine · Bowel obstruction

Introduction

Adenomyoma of the gastrointestinal tract is uncommon, characterized histologically by abnormal glandular formations lined with columnar epithelium and surrounded by smooth muscles.1-5 Adenomyoma rarely originates in the small intestine distal to duodenum, and accounts for only about 6.8% of benign tumors of the small intestine.6 We found 24 case reports of this entity in the published literature.2,4,5,7-24 This report describes a further case of adenomyoma in the small intestine, followed by a review of the pertinent literature.

Case Report

An 81-year-old woman was admitted to Omori Red Cross Hospital after vomiting three times with severe abdominal pain. Physical examination revealed slight abdominal distention without tenderness. She had no remarkable medical history except for dementia. The initial hematologic evaluation revealed the following values: hemoglobin, 14.2 g/dl; white blood cell count, 6500/μl; and platelet count, 249 × 10³/μl, with other values normal. Plain abdominal X-rays showed dilated loops of small intestine with air-fluid levels. Computed tomography (CT) also showed marked dilatation of the small intestine, but no evidence of the cause of the obstruction. Therefore we inserted a long tube, and an X-ray of the small intestine using gastrografin showed that the obstruction was mechanical, caused by a tumor of the small intestine (Fig. 1).

Laparotomy revealed a hard mass, approximately 2 cm in diameter, located about 160 cm distal to the Treitz ligament (Fig. 2). We performed a segmental resection of the jejunum with an end-to-end anastomosis. The patient had an uneventful postoperative course and was discharged on postoperative day 22.

Pathological Findings

Grossly, the mass was a 2.0 × 1.5-cm-sized lesion covered with normal mucosa (Fig. 3A,B). Paraffin-embedded blocks and sections were prepared in the conventional manner and stained with hematoxylin–eosin. Sections of the tumor revealed that it was located within the submucosa and muscularis propria, and was composed of proliferative glandular structures of variable size and irregular muscular bundles surrounding glandular elements (Fig. 3C,D). No ectopic pancreatic acini or islets were found in the lesion. Pathological examination revealed marked dilatation of the proximal intestine. The tumor cells were immunohistochemically positive for CK-7 and negative for CK-20. Some glands expressed carbohydrate antigen 19-9 and others did not. Goblet cells stained with Alcian blue were also present (Fig. 4). These findings were compatible with a diagnosis of adenomyoma of the jejunum.
Discussion

Only about 3%–6% of gastrointestinal tumors arise in the small intestine, and adenomyoma of the small intestine is extremely rare. Because of its histological characteristics, namely, abnormal glandular formations lined by columnar epithelium and surrounded by smooth muscles, it is also known as hamartoma, myoepithelial hamartoma, adenomyomatous hamartoma, foregut choristoma, and ectopic pancreas. The pathogenesis of this tumor is not well defined, but it is generally considered to be a form of hamartoma or a pancreatic heterotopia. To our knowledge, only 24 case reports have been published since Clarke’s report.