CASE REPORT Print ISSN 2234-2400 / On-line ISSN 2234-2443
Clin Endosc 2012;45:425-427 http://dx.doi.org/10.5946/ce.2012.45.4.425

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A Case of Duodenal Duplication Cyst Manifested by Duodenal Polyp

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Duodenal duplication cyst is a rare anomaly, totaling only 4% to 12% of gastrointestinal duplications, and is usually encountered during infancy or in early childhood. Most are commonly located posterior to the first or second portion of the duodenum. Presenting signs and symptoms include vomiting, decreased oral intake, periumbilical tenderness, abdominal distention, obstructive jaundice, acute pancreatitis, and gastrointestinal bleeding. The traditional treatment of a duodenal duplication cyst has been complete surgical resection, but very few cases of endoscopic treatment have been reported in the literature. Here, we report a case of duodenal duplication cyst that was manifested by a duodenal polyp.

Key Words: Duodenum; Duplication cyst; Endoscopic resection

INTRODUCTION

A duplication cyst is a spherical or tube-shaped structure covered with luminal epithelial cells, which is attached to a portion of the gastrointestinal tract. Although it is a rare congenital anomaly, it can occur anywhere in the digestive tract. It has been reported to be particularly common in the small intestine, followed by the terminal ileum, the jejunum, and the duodenum, in that order.[1-3] The incidence in the duodenum is approximately 4% to 12% or approximately one in 100,000 individuals, preferentially in the bulb and the second portion of the duodenum.[4-6] In most cases, it occurs during infancy, and is asymptomatic until its detection, although it is sometimes detected in adults. The time of manifestation of symptoms is diverse, from infancy to childhood or adulthood. The clinical features are duodenal obstruction, obstructive jaundice, and hemorrhage; and pancreatitis may develop in rare cases.[6,7] In many cases, diagnosis and treatment via cyst removal are performed simultaneously. The authors report here a case of incidental detection of a duodenal duplication cyst manifested by a duodenal polyp.

Received: December 6, 2011 **Revised:** February 27, 2012
Accepted: March 5, 2012

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CASE REPORT

A 62-year-old female visited our hospital due to a polypoid lesion in the duodenum on the health check-up endoscopy. The patient's history of diabetes, hypertension, and surgery was not surfaced earlier, and there were no special findings from her and her family's social history. On physical examination at admission, vital signs were stable and other tests were normal.

Peripheral blood test showed white blood cell 6,080/mm^3, hemoglobin 9.9 g/dL, hematocrit 29.9%, and platelets 223,000/mm^3. On biochemical analysis, the blood urea nitrogen was 23.5 mg/dL, creatinine 0.6 mg/dL, amylase 94 U/L, lipase 39 U/L, total protein 6.4 g/dL, albumin 3.8 g/dL, total bilirubin 0.34 mg/dL, direct bilirubin, 0.08 mg/dL, AST/ALT 19/22 U/L, and alkaline phosphatase 173 IU/L. The results of the chest and abdominal radiological examinations were normal.

On endoscopy, about 1-cm sized, Y-III polypoid lesion was seen at the posterior wall of the superior duodenal angle, which was soft and easily movable by pressing with a biopsy

forceps (Fig. 1A). Endoscopic ultrasound (EUS) showed that the lesion was an anechoic cystic lesion located in the third layer, and that the cyst wall consisted of three layers (Fig. 1B). For the diagnosis and treatment, endoscopic mucosal resection (EMR) was performed (Fig. 1C-E).

In the pathology of the resected polypoid lesion, the lesion was diagnosed as a duodenal duplication cyst. The cyst wall was contiguous with the duodenum and surrounded by smooth muscle layer, inner and outer surfaces of which were formed by the normal duodenal mucosa (Fig. 2). Now, 3 years after the EMR, the lesion has not recurred, and the patient is still under follow-up observation.

DISCUSSION

Gross et al.[8] defined a gastrointestinal duplication cyst in 1952 as satisfying the following three criteria: first, the cyst should be closely attached to the corresponding organ; second,

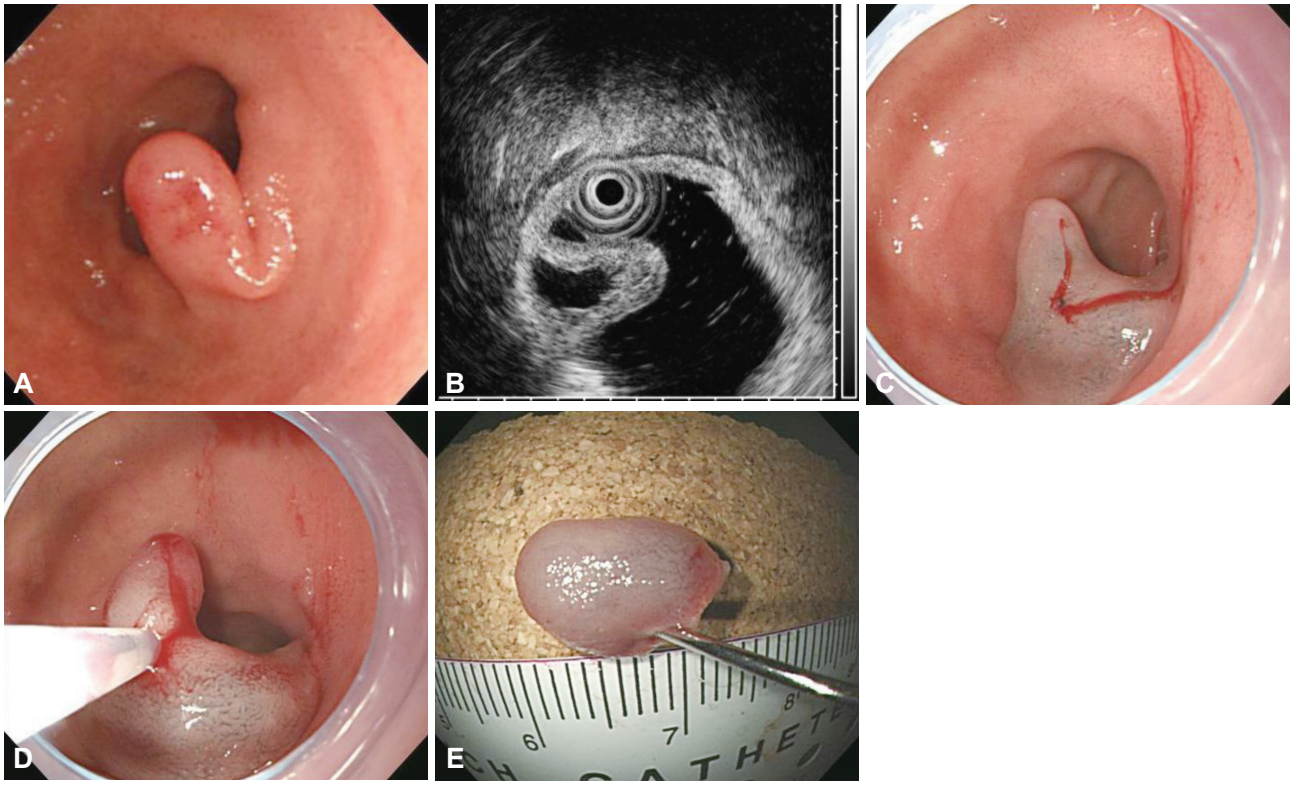

Fig. 1. Endoscopic mucosal resection of the duodenal duplication cyst. (A) A polypoid lesion at the posterior side of superior duodenal angle. (B) Endoscopic ultrasonography shows an anechoic homogenous, oval lesion originating from the submucosal layer of the duodenum wall; the wall of the cystic lesion is shown as a three-layer structure. (C) Injection of saline with indigo carmine into the submucosa. (D) After the submucosal injection, the snare is closed to capture the lesion. The lesion is then resected with a standard snare excision technique. (E) The lesion is completely removed.

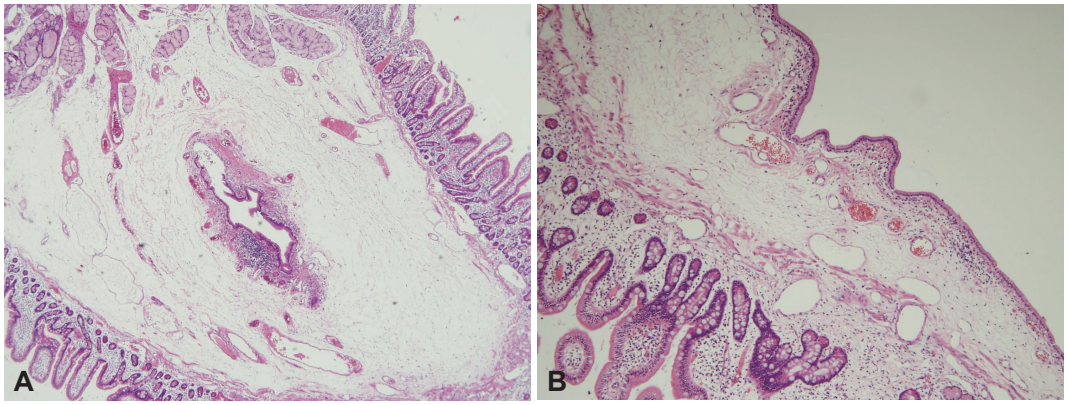

Fig. 2. Histopathologic features of the resected specimen. (A) Pathologic examination reveals that the lesion has cystic structure (H&E stain, ×40). (B) The cystic space is lined by columnar epithelial mucosa and has its own muscle layer (H&E stain, ×100).

the inside and outside of the cyst should be covered with the intestinal mucosa; and third, the cyst should have a smooth muscle layer. Histologically, duplication cysts refer to cysts that are filled with fluid and formed by well-developed smooth muscle layers and the mucosal layer, which is made up of tissues identical to the host gastrointestinal tract. Occasionally, the cysts may contain gastric or duodenal epithelial cells, or pancreatic or respiratory epithelial cells.

Although the duplication cyst is a rare congenital anomaly, it may occur anywhere along the digestive track. Its incidence in males is identical to its incidence in females.[2,3]

Most duodenal duplication cysts are filled with clear mucosal secretion, and range from 1 to 25 cm in size, depending on the volume of their secreted materials. Duodenal duplication cysts occur primarily along the anterior wall.[9]

The clinical features of duodenal duplication cysts range from non-specific symptoms such as vomiting, anorexia, abdominal pain, and abdominal distention, to small bowel obstruction due to the filling of the cyst with secretion. In addition, duodenal bleeding caused by ulceration, peritonitis, pancreatitis, jaundice, intussusception, and infection has been reported.[10-12] Diseases that should be differentiated from duodenal duplication cysts are all diseases that may appear as duodenal ulcer on the upper gastrointestinal series, such as lipoma, tumors in the ampulla of Vater, carcinoids, pancreatic pseudocysts, and choledochal cysts.[6]

Duodenal duplication cysts are occasionally detected with recurrent pancreatitis or idiopathic gastrointestinal hemorrhage. Nonetheless, few cases of asymptomatic duodenal duplication cysts, as in this case, which were incidentally detected as polypoid lesions, have been reported. Duodenal polypoid lesions are diverse; endoscopic biopsy and EUS are sometimes performed for more information in some lesions, but accurate diagnosis is ultimately confirmed by resection.

We decided to resect the lesion because of the patient's request and in order to confirm the diagnosis. Although "watchful waiting" is the typically recommended strategy for benign cystic lesion in small intestine, this approach imposes a tremendous emotional burden on patients, who can become preoccupied with thoughts that the lesion might be malignant or that complications such as bleeding or obstructions will develop.

Recently, health check-up endoscopy is very popular and so it is deemed that more cases of asymptomatic duodenal duplication cysts will be reported.

The duodenal duplication cyst is a relatively rare disease. It may be diagnosed incidentally or with abdominal pain, pancreatitis, or hemorrhage symptoms-in other words, due to associated complications. Recently, with the increase in the number of endoscopic examinations, numerous lesions are being detected incidentally. In this paper, a case of a duodenal duplication cyst which was detected as an asymptomatic polypoid lesion in an adult is reported with a literature review.

Conflicts of Interest

The authors have no financial conflicts of interest.

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