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A case of asymptomatic ileal duplication cyst associated with acute appendicitis

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ABSTRACT

Duplications of the alimentary tract are infrequent anomalies. They are most frequently located in the terminal ileum, and majority of them became symptomatic before the age of 2. Presenting symptoms may include abdominal mass, intestinal obstruction, intussusception, rectal bleeding, and abdominal pain. Preoperative diagnosis is usually difficult, intra-abdominal duplications are usually diagnosed during surgical explorations of above complications. We presented a 12-year-old girl with asymptomatic ileal duplication cyst associated with non-complicated acute appendicitis, whose imaging studies at admission were compatible with complicated perforated appendicitis.

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1. Introduction

Duplications, which rare anomalies of the gastrointestinal tract, may be located in any part of the gastrointestinal system from the oral cavity to the anus. They are most frequently seen in the terminal ileum, the ileocecal region, and the esophagus. Sixty-seven percentages of duplications develop symptoms before the age of 1, and 85% before the age of 2. The rest may remain silent until older ages or even adulthood [1,2]. Symptoms usually include intra-abdominal mass, rectal bleeding, intestinal obstruction, abdominal pain, and intussusception [3]. Preoperative diagnosis of GIS duplications is usually difficult. Radiologic imaging studies may not be enough for definitive diagnosis. We present a case of asymptomatic ileal duplication cyst associated with uncomplicated acute appendicitis.

2. Case report

A 12-year-old girl was referred to our pediatric surgery department complaining of abdominal pain, nausea, and vomiting. On physical examination the patient had abdominal tenderness and guarding in the right lower quadrant. Laboratory studies showed leukocytosis (WBC: 13.51 10^9/L), and increased C-reactive protein level (33 mg/dl). Abdominal X-ray did not demonstrate any pathologic finding. The abdominal ultrasonography (US) showed an inflamed appendix exceeding 7 mm in diameter and an adjacent heterogenous fluid collection measured approximately 6 × 4.5 cm. The patient was operated with the preliminary diagnosis of acute complicated appendicitis. Surgical exploration revealed an acute suppurative non-complicated appendicitis, and a 7 × 6 cm non-complicated duplication cyst with a common wall with the ileum 4 cm proximal to the ileocecal valve (Fig. 1). Appendectomy, segmental ileal resection including the duplication cyst, and end-to-end ileal anastomosis was performed. Following surgery we questioned the parents for detailed past medical history, any previous chronic abdominal pain, vomiting, lower GIS bleeding were noted. The patient was started on enteral feeding on postoperative day 5, and discharged without any complications. Histopathology examination of the specimens showed appendicitis with local peritonitis, and duplication cyst lined with ileal mucosa without any ectopic tissue (Fig. 2).

3. Discussion

Gastrointestinal duplications are exceptional congenital anomalies which can be seen anywhere in the gastrointestinal tract from the mouth to the anus. Its prevalence is believed to be between

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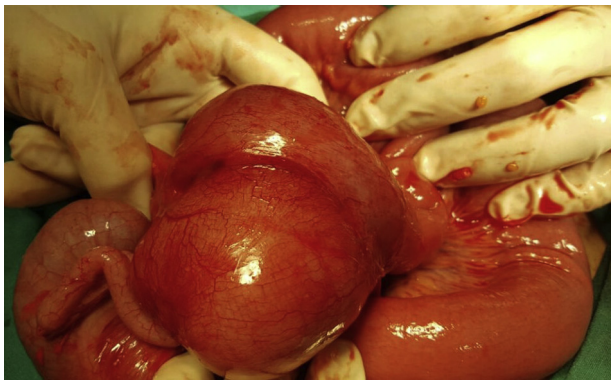

Fig. 1. Operative image of ileal duplication cyst.

1:4500–1:10000 [1,2]. Theories such as persistence of fetal bowel diverticula, defect in recanalization of the solid phase of the primitive gut, partial twinning, and split notochord theory are suggested in its etiology. Although its cause is not known clearly, the most frequently accepted theory is "the intrauterine vascular accident theory" [3]. Whilst this developmental anomaly is most commonly seen in the ileum, it may be seen in the esophagus, duodenum and rectum. Duplication has three macroscopic types: cystic, tubular and intramural. Cystic duplication is the most common type [4,5]. Duplication cysts have a smooth muscle layer in the cyst wall, and the cyst is lined with the mucosa of adjacent alimentary tract, which may contain ectopic digestive tract tissue such as gastric mucosa or pancreatic tissue [6,7]. In our case, the duplication was detected in the ileum, where it is most commonly seen, and was a cystic duplication, which is the most common type, and was lined by ileal intestinal epithelium along with small number of tubular structures beneath the mucosa and a common muscle layer with the adjacent intestinal tissue. However heterotopic tissue was not observed in our case.

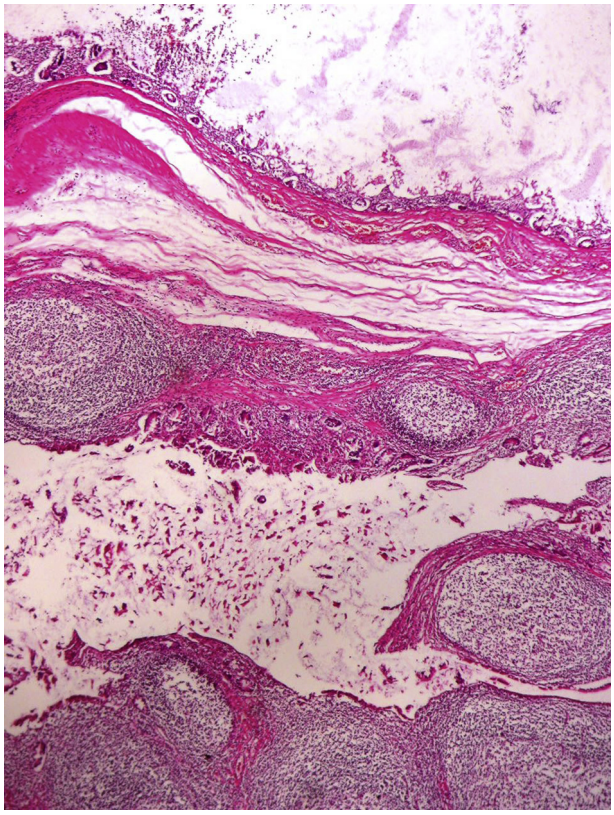

Fig. 2. The ileum wall where the Peyer's patches are observed and the duplication cyst wall lined by ileal mucosa, which has a common muscular wall with the ileum, H&E,100×.

Although most of the duplications become symptomatic before age 2, a few may remain asymptomatic as seen in our patient. Clinical symptoms of duplication cysts may vary depending on its localization, size, and whether it contains heterotopic mucosa or not. Symptoms usually include intra-abdominal mass, rectal bleeding, intestinal obstruction and abdominal pain [8,9]. Abdominal pain may be due to volvulus, bowel obstruction associated with intussusception, or perforation related to ulceration caused by ectopic gastric mucosa. In our case, clinical symptoms were abdominal pain, nausea, and vomiting, but these symptoms were related to the accompanying suppurative appendicitis since the ileal duplication was non-complicated. The duplication cyst was detected asymptomatically and incidentally, as well ass adenocarcinoma of ileal duplication cysts have been reported in adulthood [3,10].

Duplication cysts are usually difficult to diagnose preoperatively due to their wide spectrum of signs and symptoms. The most widely used imaging methods in its diagnosis include US and contrast studies with barium. Computed tomography and magnetic resonance imaging are less commonly required. Today, a significant number of duplication cysts are diagnosed during prenatal imaging studies [9]. In US, duplication cysts are identified with the presence of the echogenic inner mucosal layer and hypoechoic outer muscular layer [11]. US imaging of our case showed an inflamed appendix exceeding 7 mm in diameter, and adjacent heterogeneous fluid collection of approximately 6 × 4.5 cm, and accordingly the acute complicated appendicitis was considered as the preliminary diagnosis. However, this finding obtained on the preoperative US imaging was related to duplication cyst.

Duplication cysts are infrequent congenital intestinal anomalies. The majority of cases become symptomatic, and diagnosed before the age of 2; they may also be encountered asymptomatically and incidentally with acute appendicitis in later ages, as in the case presented here. Even when cystic duplications are detected incidentally, the suggested treatment is surgical excision due to potential complications such as volvulus, obstruction, invagination and bleeding.

Conflict of interest

None.

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