

An ileal duplication cyst manifested as an ileocolic intussusception in an adult

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ABSTRACT

An enteric duplication cyst presenting as enterocolic intussusception is an exceptional clinical entity. We herein report a rare case of an ileal duplication cyst that manifested as an ileocolic intussusception. A 19-year-old woman was hospitalized due to right upper quadrant pain. Colonoscopy revealed a polypoid mass protruding into the colonic lumen. Ultrasonography demonstrated intussusception with a teardrop-shaped cystic mass at the tip. Computed tomography also showed ileocolic intussusception with a 2.5 cm-sized round cystic mass at the tip of intussusceptum. Microscopically, the cystic wall consisted of a well-defined smooth muscle coat and heterotopic gastric mucosa, consistent with an enteric duplication cyst. This case highlights an ileal duplication cyst as an uncommon cause of adult ileocolic intussusception. To the best of our knowledge, this is the first case of enteric duplication cyst identified as a pathological lead point for enterocolic intussusception in an adult. Enteric duplication cysts should be included in the differential diagnosis of cystic-leading lesions for adult intussusceptions.

Keywords: Enteric duplication cyst, enterocolic intussusception, adult

INTRODUCTION

Intussusception occurs when a portion of the digestive tract becomes telescoped into the adjacent bowel segment. This condition usually occurs in children between 6 months and 2 years of age. In the past, intussusception was a severe condition with high morbidity and mortality rates. Currently, prompt diagnosis and effective treatment lead to a favorable outcome in most cases. Enteric duplication cysts are an uncommon congenital abnormality that manifest before 2 years of age in 80% of cases and that can occur anywhere along the gastrointestinal tract on the mesenteric side. The presenting signs and symptoms of enteric duplication cysts include abdominal distension, vomiting, bleeding, a palpable abdominal mass and urinary infrequency. The clinical manifestations are usually intestinal obstruction from adjacent pressure, volvulus and rarely intussusception (1). A thorough review of the literature revealed a few case reports of enteric duplication cysts causing enterocolic intussusception in infants (2-5); however, no such cases have been reported in adults to the best of our knowledge. We herein report a rare case

of an ileal duplication cyst manifested as an ileocolic intussusception in a young adult woman.

CASE PRESENTATION

A 19-year-old woman without medical or surgical history was hospitalized due to right upper quadrant pain. Physical examination revealed a tender palpable mass in the right upper quadrant of the abdomen. Laboratory results were within the normal range. Colonoscopy revealed a polypoid mass protruding into the colonic lumen (Figure 1a). Ultrasonography (US) showed intussusception with a teardrop-shaped cystic mass at the tip, which had a double-layered wall consisting of a hypoechoic outer layer and an echogenic inner layer (Figure 1b). Computed tomography (CT) demonstrated ileocolic intussusception with a cystic mass at the tip of the intussusceptum (Figure 1c). A normal-appearing appendix was not included in the intussusception. The cystic mass was considered to act as a lead point for the intussusception. Exploratory laparotomy showed an invagination of the terminal ileum and mesentery into the cecum through the ileocecal valve. Due to the

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impossibility of reducing the intussusception, a right hemicolectomy was performed (Figure 2a). Grossly, the resected specimen demonstrated a round cystic lesion extending from the terminal ileum into the colon measuring 2.5 cm in diameter (Figure 2b). Microscopically, the cystic wall consisted of a well-defined smooth muscle coat and heterotopic gastric mucosa (Figure 2c), consistent with an enteric duplication cyst. The patient remained well without specific events or complications for 2 years postoperatively.

DISCUSSION

The lead point for enterocolic intussusception in adults can be located in the small intestine, colon or appendix, and a wide variety of lesions in the ileum may be responsible for ileocolic intussusception. Benign tumors including lipoma, inflammatory fibroid polyp and hamartomatous polyp as well as malignant tumors such as lymphoma and ileal cancer, and Meckel's diverticulum have all been described as lead points for ileocolic intussusception (6). In our interesting case, the lead point was an enteric duplication cyst in the terminal ileum. Enteric duplications are cystic or tubular structures attached to the mesenteric side of the intestine, often sharing a common smooth muscle wall and vascular supply with histological features of a layered smooth muscle coat and gastrointestinal mucosal lining (7).

Both intussusception and duplication are unusual in adult patients, and the combination of both events in one patient is very rare. Although there have been 4 reported cases of enteric duplication cysts that manifested as enterocolic intussusceptions, all the patients in these cases were neonates or infants under 2 years of age (2-5). To the best of our knowledge, this is the first adult case of an enteric duplication cyst identified as a pathological lead point for enterocolic intussusception.

Intussusception in adults is estimated to account for only 5% of all intussusceptions and causes only 1% of bowel obstructions (8). However, about 90% of intussusceptions in adults are caused by a definite underlying disorder such as a neoplasm or postoperative condition (9). Thus, a correct and timely diagnosis is not only necessary to resect the underlying lesion that serves as the lead point but also to avoid the complications of bowel infarction and perforation secondary to obstruction. CT is the most sensitive diagnostic modality used to image intussusceptions. On CT analysis, intussusceptions reveal a pathognomonic appearance of a complex soft tissue mass consisting of the outer intussusciens and the central intussusceptum. When the CT beam is parallel to the longitudinal axis of the intussusception, it appears as a "sausage-shaped" mass; when the beam is perpendicular to its axis, it appears as a "target"

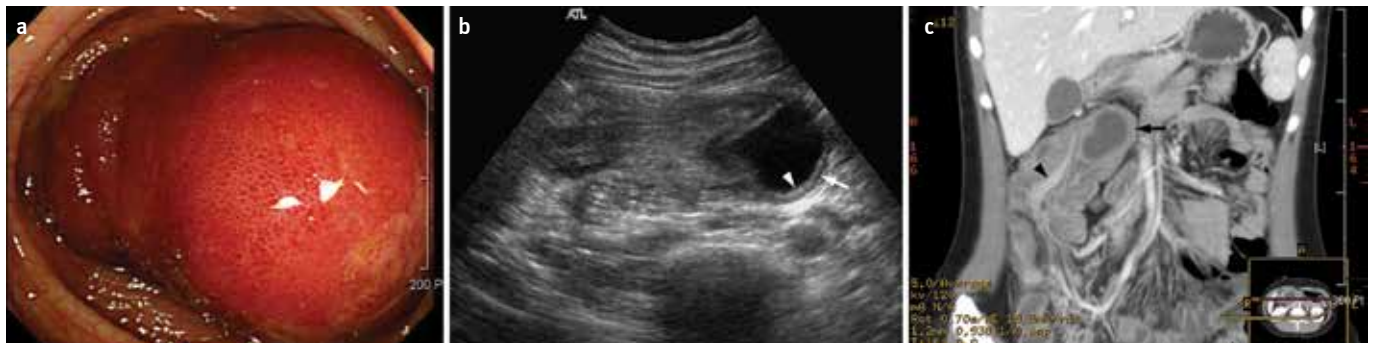


Figure 1. a-c. Imaging findings. Colonoscopy reveals a polypoid lesion protruding into the colonic lumen (a). Longitudinal US of the right upper abdomen demonstrates an echogenic lesion at the tip of the intussusceptum. The cystic mass shows a hypoechoic outer muscular layer (white arrow) and an echogenic inner mucosal layer (white arrowhead), suggestive of an enteric duplication cyst (b). Coronal reconstructed CT images shows a sausage-shaped mass with a well-enhanced portion in the ascending colon, representing the bowel wall of the intussusciens within the intussusceptum at its periphery and a central fatty density, representing mesenteric fat. The enteric duplication cyst in the terminal ileum (black arrow) acts as a lead point for the ileocolic intussusception. Linear enhancing structures (black arrowhead) are mesenteric blood vessels (c).



Figure 2. a-c. Gross and microscopic findings. The specimen from a right hemicolectomy shows an invagination of the terminal ileum into the cecum (white arrowheads) (a). A round cystic lesion measuring 2.5 cm in diameter extends into the colonic lumen through the ileocecal valve (b). Microscopically, the cystic wall is enclosed by a well-defined smooth muscle wall and is lined with heterotopic gastric mucosal epithelium (Original magnification x100; Hematoxylin-eosin stain) (c).

mass (10). In contrast, the most common imaging modalities used to image enteric duplication cysts is US. The presence of a double wall or muscular rim is well described in the literature, which refers to the appearance of a cyst mimicking the gastrointestinal tract with a hypoechoic rim of tissue representing the smooth muscle layer and an echogenic inner margin corresponding to the mucosa (11). Identification of this trait on an US of an abdominal mass has been regarded as characteristic of an enteric duplication cyst (12,13).

Recognition of an intussusception in an adult patient may often be difficult and may represent a major challenge for inexperienced clinicians due to the importance of prompt surgical treatment. Its association with other congenital bowel abnormalities may be even unfamiliar to radiologists or clinicians. The differential diagnosis of an enteric duplication cyst in association with enterocolic intussusception primarily includes omental cyst, lymphangioma, mesenteric lymphoma, fluid encapsulated in the mesentery during intussusception, intramural neoplasm and Meckel's diverticulum. It is important to be aware of the imaging spectrum and the clinical features of intussusception secondary to congenital conditions because imaging plays a crucial role in the diagnosis and management of these patients.

In summary, we report a rare case of an ileal duplication cyst manifested as an ileocolic intussusceptions in a young adult woman. This case highlights the ileal duplication cyst as an uncommon cause of adult ileocolic intussusception. Enteric duplication cysts should be included in the differential diagnosis of cystic-leading lesions for intussusceptions in adults.

Conflicts of Interest: No conflict of interest was declared by the authors.

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