Letter to the Editor

Unusual radiological and endoscopic findings of a small intestinal duplication in an adult

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Introduction

Initially reported in 1897, alimentary tract duplication (ATD) is a rare congenital malformation that can affect any segment along the digestive tract. It occurs most commonly in the ileum of infants but rarely emerges in adults (1). The preoperative diagnosis of this disease poses significant challenges due to its diversified clinical presentations and intricate anatomical structure. Herein, we present the clinicopathological, radiological, and endoscopic findings of a tubular intestinal duplication located inside the lumen of the ileum in a young man. Our objective is to raise the awareness among clinicians and radiologists of this rare condition in adulthood.

Case presentation

A 24-year-old young man attended our hospital due to experiencing melena 1 week prior and hematochezia over the previous 4 days. He reported no other discomfort, such as nausea, vomiting, fever, or changes in bowel movements. He further denied any previous history of malignancy or abdominal surgery. The physical examination elicited slight tenderness in the right abdomen, without rebound pain or a palpable abdominal mass. Routine blood tests showed a reduced hemoglobin level (53 g/L; reference range, 130–175 g/L). Tumor markers and liver and renal function indices were within normal ranges. Routine gastroscopy and colonoscopy at a local hospital before admission revealed no sources of bleeding. Computed tomography (CT) yielded "sausage" and "doughnut" signs in the small bowel in the right abdomen (*Figure 1A*,*1B*) suggestive of intestinal intussusception, with an intraluminal focal fat-dense nodule. Double-balloon enteroscopy (DBE) examination via the anal route was performed and revealed a polypoid lesion with ulcerous areas in the ileum, approximately 100 cm proximal to the ileocecal valve (*Figure 1C*). Subsequently, a laparoscopic partial small bowel resection with a side-to-side anastomosis was executed. Intraoperative opening of the specimen showed a 12×2 cm tubular lesion within the lumen, originating from the mesentery-side wall of the native ileum (*Figure 1D*). Pathological analysis confirmed the diagnosis of intestinal duplication, characterized by the presence of intestinal mucosa, irregularly arranged muscular layers in the tubular wall, and hyperplasia of fatty tissue in the distal segment of the lesion (*Figure 1E*,*1F*). The patient experienced a smooth recovery and was discharged on the 11th postoperative day. During 1 year of follow-up, there were no signs of gastrointestinal bleeding or any other discomfort. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is

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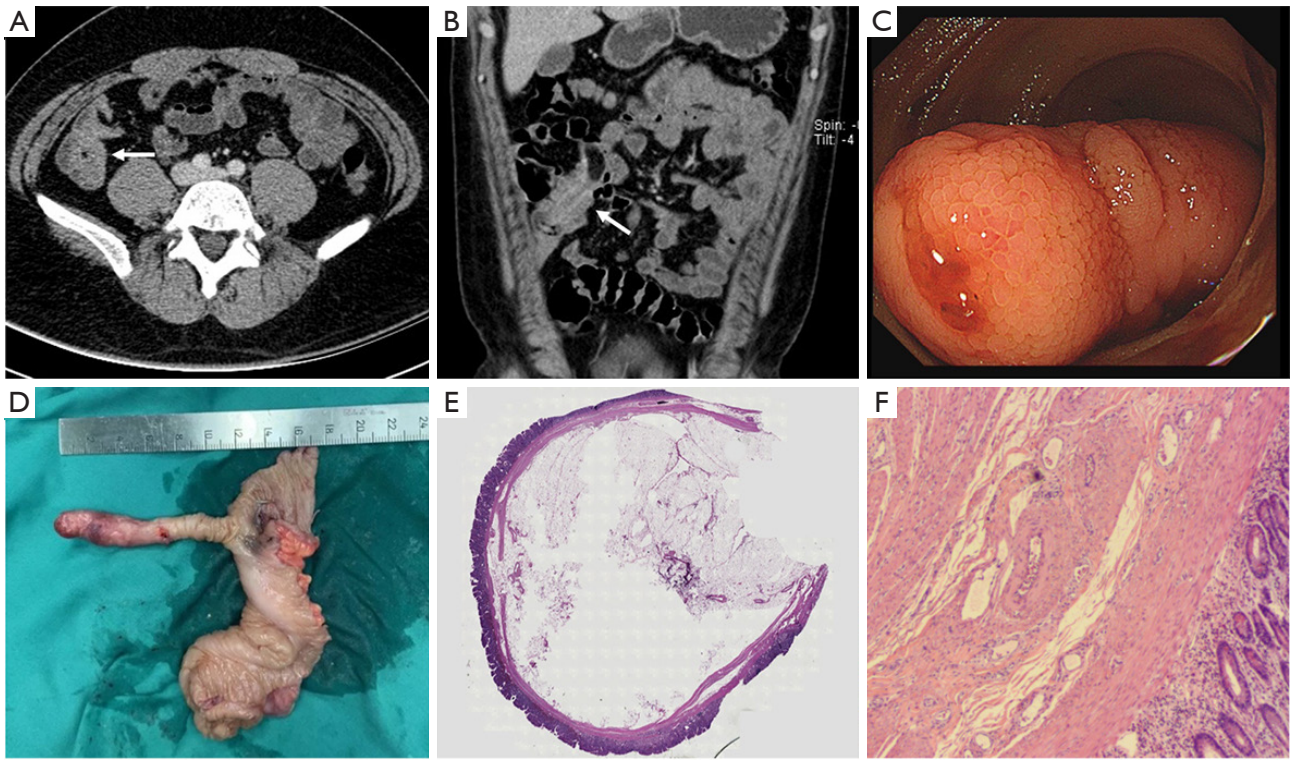

Figure 1 CT (A,B), DBE (C), and pathological (D-F) findings of the case. Axial (A) and coronal (B) views of contrast-enhanced CT respectively showed the "doughnut" and "sausage" signs in the small intestine of the right abdomen (white arrows). DBE revealed a polypoid lesion with ulcerous areas in the ileum (C). Examination of the surgical specimen indicated a communicating intraluminal tubular lesion measuring approximately 12×2 cm in size (D). The pathological section of the distal segment (E) revealed a duplicated intestinal wall covering regions of adipose tissue hyperplasia (HE; original magnification 5×). The wall of the tubular duplication (F) exhibited intestinal mucosa and irregularly arranged muscular layers (HE; original magnification 200×). CT, computed tomography; DBE, double-balloon enteroscopy; HE, hematoxylin and eosin staining.

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Discussion

Duplication of the digestive tract is infrequent (1/10,000 in newborns), and its diagnosis in adulthood is extremely rare (1). It occurs most commonly in the ileum, followed by the colon, esophagus, stomach, and duodenum. The mechanism of its emergence is not entirely clear. Several hypotheses have been posited and include split notochord theory, partial twinning, persistence of embryonic diverticula, recanalization defects, intrauterine vascular insult, and environmental factors (2). Duplications can be categorized into cystic (80%) and tubular (20%) types based on anatomical structures. They may or may not communicate with the adjacent alimentary tract and manifest asymptomatically or present diverse, nonspecific abdominal symptoms. In adults, ATD is an atypical source of gastrointestinal bleeding, with hematochezia noted as the initial symptom in 10.5% of cases (3). A study in Japan reported that only approximately 10% of the examined cases were accurately diagnosed before surgery (3).

Various diagnostic methods have proven effective in investigating ATD in adults and primarily included gastrointestinal radiography, ultrasonography (US), abdominal CT, magnetic resonance imaging (MRI), and endoscopy. Barium meal radiography can diagnose ATD only when it communicates with the native intestinal lumen. US is a commonly used imaging method in diagnosing abdominal ATD, especially in pediatric and prenatal diagnosis. Classical US findings of ATD include a cystic structure adjacent to the gastrointestinal tract and the presence of cyst wall peristalsis during dynamic observation (4). The cystic lesion typically manifests as

Table 1 Summary of reported cases of ATD with similar radiological and endoscopic findings

Author, year	Age (years)	Gender	Location	Clinical manifestation	CT or MRI findings	Endoscopic findings
Kim, 2014 (29)	19	F	Ileum	Right upper quadrant pain	A sausage-shaped mass suggesting ileocolic intussusception with a cystic mass at the tip	A polypoid mass protruding into the lumen
Kyo, 2016 (30)	20	M	Colon	Intermittent right flank pain	Colon intussusception with a cystic mass	A large submucosal mass
Al-Shaibi, 2019 (31)	24	M	Colon	Recurrent periumbilical pain	Ileocolic intussusception with a rounded lesion as a lead point	A large hyperemic submucosal mass
Zhang, 2021 (32)	31	F	Ileum	Intermittent hematochezia	"Doughnut" sign, suggesting intussusception of the ileum	A protruding lesion in the lumen

ATD, alimentary tract duplication; CT, computed tomography; MRI, magnetic resonance imaging.

a "double-wall" sign, comprising a hyperechoic mucous layer and a hypoechoic muscular layer. Cross-sectional imaging techniques, typically CT and MRI, can provide comprehensive information of the location, scope, and complications, while the associated abnormalities and relationship with the adjacent structures can be discerned through three-dimensional imaging. Therefore, these methods are crucial for the pre-treatment evaluation of ATD. Meanwhile, endoscopic technology, particularly capsule endoscopy and DBE, have been deemed valuable for diagnosing various anomalies of alimentary tract. In adults with digestive symptoms, a combined method of imaging and endoscopic techniques is often employed to identify the underlying etiology. We reviewed prior reported cases of ATD with both positive radiological (CT or MRI) and endoscopic findings in the literature. The typical features of ATD are as follows: on CT or MRI, it manifests as an abnormal cystic or tubular structure adjacent to the digestive tract (5-25), while on endoscopy, it is characterized by a bifurcated or diverticular-like lumen (5-15) or by extrinsic compression of the digestive wall via a bulging mass (16-25). In the minority of ATD cases accompanied by malignancy or inflammation, the presentation may be atypical and diverse and include, for example, irregular thickening of the gastrointestinal wall or a complex soft tissue mass on radiological imaging and luminal stenosis or a cavity-forming mass on endoscopic examination (26-28). In rare instances, ATD may display the "sausage" and "doughnut" signs mimicking intussusception on CT or MRI; meanwhile, it may exhibit an intraluminal protruding structure on endoscopy (29-32) (*Table 1*), as observed in the case reported here. This distinctive manifestation typically arises from localized enteric intussusception induced by a cystic duplication (29-31) or on even rarer occasions—as in our case—a tubular duplication located within the intestinal lumen (32). When radiological signs resembling intussusception emerge in an adult, the primary concern often revolves around the association with an underlying malignancy or inflammatory bowel disease. Nevertheless, further examinations, including endoscopy, are required to exclude alternative etiologies, such as gastrointestinal congenital anomalies similar to ATD, especially in young adults.

Although there are no specific guidelines on managing ATD, surgical resection of the lesion with the adjacent intestine is generally advocated for definitive diagnosis, relief of symptoms, and prevention of further potential complications and malignancies (33).

Intestinal duplications with delayed onset of digestive symptoms in adults, particularly the tubular subtype, are fairly uncommon. Despite being challenging, a combination of radiological and endoscopic approaches may facilitate the preoperative diagnosis of such malformations. Surgical intervention persists as the most important treatment for alleviating symptoms and reducing the potential risks of serious adverse outcomes.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-1872/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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