Case Report

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Tubular colonic duplication in an adult: case report and brief literature review

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Abstract

Gastrointestinal tract duplication is a rare congenital anomaly that can occur anywhere along the alimentary tract. Most of the reported patients present with acute abdomen during childhood. We describe a case of tubular colonic duplication in an adult. The patient was a 25-year-old woman who presented with abdominal pain, bloating, nausea, and emesis for 3 days. The physical examination was remarkable for abdominal distension, tenderness, and rigidity. Abdominal computed tomography scan revealed abnormal intestinal dilatation. Exploratory laparotomy was performed, and tubular colonic duplication was identified intraoperatively. The diagnosis was verified by postoperative pathology results. The patient was discharged on postoperative day 14 and followed for 2 years without specific events or complications. Furthermore, we reviewed the published literature on colorectal duplication in adults for the past two decades.

Keywords

Colonic duplication, congenital anomaly, adult, abdominal distension, intraoperative diagnosis, adenocarcinoma risk

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Introduction

Gastrointestinal tract duplication is a rare congenital anomaly that can occur anywhere along the alimentary tract. Approximately 80% of duplications occur in the abdomen, with others occurring in the chest.[1] For digestive duplications, the ileum is the most common site (30%–35%) and the colon is

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the least common site (7%–20%).[2,3] The duplication can widely vary in size and shape; it is cystic in more than 80% of patients and tubular in the rest.[4,5]

Gastrointestinal tract duplication usually manifests during the neonatal period or childhood; most patients (67%–80%) are diagnosed before the age of 2 years.[2,3] Few patients remain asymptomatic until adulthood. Complications such as volvulus, bleeding, intussusception, perforation, and malignant transformation are typically the first clinical manifestations of gastrointestinal tract duplication. It is difficult to make a preoperative diagnosis. Surgery is usually performed to treat complications, and duplication is diagnosed intraoperatively in most cases.

Herein, we describe the case of a 25-year-old woman who presented with abdominal pain and was diagnosed intraoperatively with tubular colonic duplication. The rarity of this case is reflected in three aspects: adult age at symptom onset, colonic location, and tubular subtype.

Case report

A 25-year-old Han Chinese woman presented to the emergency department of our hospital with abdominal pain, bloating, nausea, and emesis for 3 days. She could pass stool and flatus but with significant reduction. She denied bloody stools, fever, chills, or weight loss. In the past decade, she had experienced abdominal pain that would disappear after rest without any treatment. There was no known precipitating factor for the abdominal pain. She reported no other symptoms, such as constipation or diarrhea. There was no significant family history of genetic disease or cancer. Her antenatal history was unremarkable. Physical examination showed abdominal distension, tenderness, and rigidity. Routine laboratory values were unremarkable. After ultrasonography revealed an expanded intestinal lumen, we performed abdominal computed tomography (CT), which showed abnormal dilatation of the intestine (Figure 1). A nasogastric tube was inserted, and approximately 300 mL of dark green fluid was drained.

Because of the peritoneal irritation and CT scan result, exploratory laparotomy was performed. The dilated intestine was intraoperatively identified as a duplicated tube volvulus of the distal part of the descending and proximal part of the sigmoid colon. No intestinal adhesions or coexistent congenital anomalies were found.

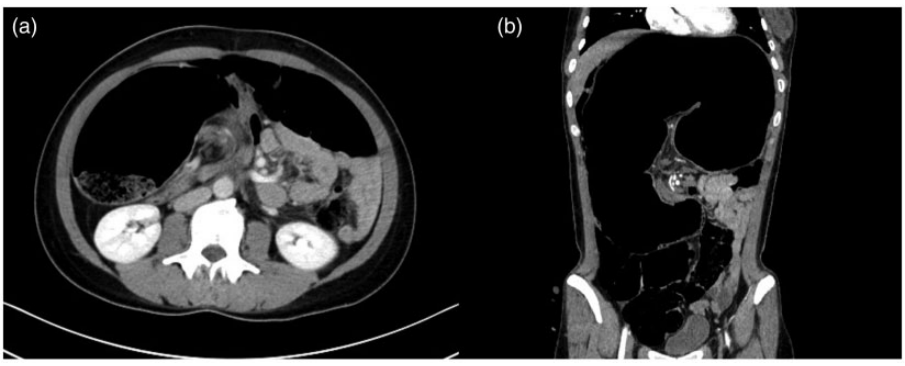

Figure 1. Preoperative computed tomography scan of the abdomen showing intestinal dilatation. (a) axial plane, (b) coronal reconstruction showing a hugely dilated intestinal tube and anomalous enteric lumen in left upper quadrant.

The anomalous segment had separate intestinal walls. There was only one connection between the anomalous and normal lumens at one end of the duplicated colon; that is, a Y-shaped tubular duplication. Partial colectomy of the descending and sigmoid colon was performed, followed by end-to-end anastomosis without ostomy. Postoperative pathology findings confirmed colonic duplication (Figure 2). Grossly, the duplicated colon was approximately 72 cm long and measured 15 cm in greatest diameter. Microscopically, the duplicated colonic segment showed muscular hypertrophy. No gastric heterotopy or adenocarcinoma was found; 10 lymph nodes were identified and all were unremarkable. The patient developed fever postoperatively, which was responsive to an antipyretic and was categorized as a grade I surgical complication according to the Clavien–Dindo classification system.[6] She was discharged on postoperative day 14 and followed for 2 years without specific events or complications.

This report was reviewed and approved by the Peking Union Medical College Hospital Institutional Review Board. The patient provided written informed consent for publication of this case report.

Discussion

Gastrointestinal tract duplication is defined by three features: (1) encircling, well-formed smooth muscular layers, (2) a mucosal layer, and (3) association with some part of the gastrointestinal tract.[7] Communication between the anomalous and actual enteric lumens may or may not be present.[8] The classification system for gastrointestinal tract duplications generally depends on the morphology and origin.[9,10] McPherson and colleagues[11] described their classification for colonic duplication in 1969 as follows: type I, simple cysts; type II, diverticula; and type III, tubular colonic duplication. This type of classification is widely used because of its simplicity.

Several theories have been proposed to explain the formation of duplications, such as aberrant luminal recanalization, embryonic diverticula, a split notochord, partial twinning, and environmental factors.[4,10,12] Thus far, no single theory has perfectly explained all types of duplications; many different factors might be involved in the formation of gastrointestinal tract duplication.

Gastrointestinal tract duplication in adults is rare and mostly described in case

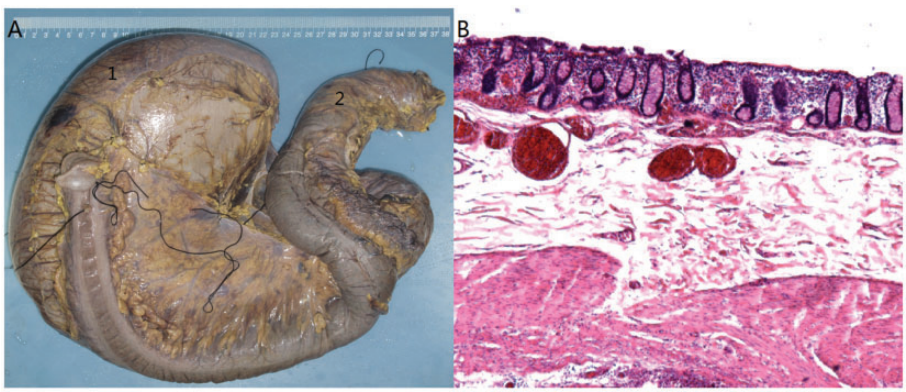

Figure 2. Postoperative pathology results. (a) Macroscopic findings of the colon specimen; (1) duplication tube, (2) native colon. (b) Microscopic histopathology of the duplicated segment. Pathological examination revealed digestive mucosa and well-formed smooth muscular layers, consistent with true duplication of the colon. Hematoxylin and eosin stain, magnification ×40.

reports. We reviewed all literature in English about colorectal duplication in adults published in the past two decades. Because of advancements in medicine, current diagnostic techniques and treatment methods are very different from those of the past decades. Therefore, we excluded cases diagnosed earlier than two decades ago. Forty-one reports with 50 cases were reviewed. The clinicopathological summary of these cases is shown in Table 1.

Adult patients' symptoms were variable and nonspecific: acute or chronic abdominal pain, an abdominal mass, bloody stools, bowel obstruction, diarrhea, and weight loss. According to our review, the most common clinical presentation was pain in the abdomen, flank, or lumbar region. The cause of this pain might be distention of the duplication or compression of the adjacent organs.[5,13,14] In our patient, all symptoms were caused by distension and volvulus of the tubular colonic duplication. Gastric mucosa exists in about 50.8% of cystic duplications,[2] so ectopic gastric mucosa ulcerations are common. This might explain hemorrhage as a symptom of digestive duplications. In our review, hemorrhage was the main symptom in three patients. It is worth mentioning that only 13 (26%) patients had a history of gastrointestinal complaints. Moreover, four (8%) patients had no gastrointestinal complaints, and the diagnosis of colorectal duplication was incidental.

It is not easy to diagnose gastrointestinal tract duplication preoperatively, and misdiagnosis is common.[2,4] The frequently used diagnostic modalities, such as plain abdominal radiography and ultrasonography, provide limited information; a gas-filled structure, for example. Useful, feasible diagnostic methods are barium enema, CT, and colonoscopy. Any of these methods can determine communication between the anomalous and actual enteric lumens. CT is the most suitable diagnostic method for detecting digestive duplication,[15] because it is easy to perform, especially in pediatric and critically ill patients. Cystic duplication can be mistaken for a cystic tumor of adjacent organs,[4,14] especially in patients in whom

Table 1. Clinicopathological summary for literature review of 50 adults with colorectal duplication*

Feature	Number
Demographics	
Mean age, years (range)	40.2 (18–76)
Male patients (%)	24 (48%)
Presenting symptoms	
Abdominal pain	23
Mass	7
None	4
Bloody stools	3
Other[a]	13
Preoperative diagnosis	
Tumor/mass/cyst	20
Duplication	19
Diverticulum	4
Other[b]	7
Site of duplication	
Transverse colon	12
Sigmoid colon	8
Cecum	8
Ascending colon	6
Rectum	5
Left colon	4
Descending colon	3
Other[c]	4
Patients with previous symptoms	13 (26%)
Patients with other malformations	7 (14%)
No luminal communication of duplication with native colon	7 (14%)
Duplication complicated by adenocarcinoma	5 (10%)
Patients who did not undergo surgery	3 (6%)

*From reports published in the past two decades.
[a]Other presenting symptoms included occlusion (n = 3), diarrhea (n = 3), distention (n = 2), constipation (n = 1), abscess (n = 1), discomfort (n = 1), weight loss (n = 1), and rectovaginal fistula (n = 1).
[b]Other preoperative diagnoses included abscess (n = 2), dilated intestinal loop (n = 2), megacolon (n = 1), inflammation (n = 1), and appendicitis (n = 1).
[c]Other sites included distal colon (n = 1), colon and rectum (n = 1), total colorectum and terminal ileum (n = 1), and entire colon and ileocecal junction (n = 1).

luminal communication does not exist. Sometimes the correct diagnosis can only be made during exploratory laparotomy. According to our review, seven patients were reported to have duplications without communication with the actual lumen. The diagnosis of duplication was identified preoperatively in only 38% (19/50) of patients. The most common misdiagnosis was a tumor, mass, or cyst, which occurred in 40% (20/50) of patients. The transverse colon was the most common site for colonic duplication.

Surgery is the treatment choice for symptomatic digestive duplications.[16] For asymptomatic patients, the management strategy is controversial;[17] however, most authors advocate surgical resection to prevent future complications and to eliminate the risk of adenocarcinoma.[4,18,19] Generally, resection of the duplication and adjacent intestine should be performed. In our case, we excised the colonic tubular duplication and segments of the descending and sigmoid colons. Only some small cystic duplications can be excised without colonic resection.[4] In our review, three patients did not undergo surgery (two because of silent clinical manifestation and one because of satisfactory drug treatment). Two of them were followed for 4 and 6 months, and no sequelae were observed. Five patients were reported to have gastrointestinal tract duplication complicated by adenocarcinoma.[20–23]

Colonic duplication can be associated with abnormalities of other organs. Sometimes a colovesical or rectovaginal fistula can coexist.[24–26] In the reports reviewed, 14% (7/50) of patients had other congenital malformations. Physicians should assess patients for other malformations when digestive duplication is diagnosed. In our patient, there was no coexisting congenital anomaly.

In conclusion, colonic duplication is a rare congenital anomaly that is usually diagnosed and treated during childhood. Some adults can harbor a silent duplication for many years until complications occur. The clinical presentation can include abdominal pain, an abdominal mass, bloody stools, bowel obstruction, and diarrhea. It is difficult to make the preoperative diagnosis of colonic duplication, and CT is the most suitable diagnostic method. The transverse colon is the most common site of colonic duplication, and surgery is advocated for symptomatic and asymptomatic patients.

Authors' contributions

Xin Wu planned the review, analyzed the data, and drafted the manuscript. Xiequn Xu planned the review and revised the manuscript for relevant content. Chaoji Zheng collected and analyzed the data and drafted the manuscript. Binglu Li collected and analyzed the data, and revised the manuscript for relevant content. All authors read and approved the final manuscript.

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The authors declare that there is no conflict of interest.

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