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Congenital Tubular Duplication of Colon in an Adult Presented by Abdominal Pain and Constipation: A Case Report and Review of the Literature

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Abstract

Introduction: Congenital duplications of the intestinal tract are rare diseases, observed mainly in ileum and stomach, while less than 7% of cases occur in the colon. They are mainly diagnosed during early childhood, while few cases may remain asymptomatic until adulthood.

Case Report: We present a 31-year-old female with chronic abdominal pain without significant past medical history. Pre-operative diagnosis was an enterocolic fistula, which was considered after colonoscopy. Abdominal surgery with midline incision was performed for the patient with suspicion of congenital colon anomaly. During the surgery, the surgeon found an additional colon closed loop that originated from the posterior aspect of the transverse colon and was attached posteriorly to the sigmoid colon. Microscopic examination revealed tubular duplication of the colon with marked dilatation, focal mucosal ulceration, granulation tissue, and mild chronic nonspecific inflammation.

Discussion: As congenital duplication of colon is a rare condition with non-specific clinical symptoms, high suspicion of the physician and thorough examination can help the diagnosis and surgical treatment as soon as possible to stop patients' pain and associated problems.

Keywords: Abdominal Pain, Colonic Diseases, Congenital Abnormalities, Constipation

1. Introduction

Multiple congenital abnormalities may occur in the Gastrointestinal (GI) tract, while structural anomalies, including developmental defects, anomalies of rotation and fixation, duplications, and anomalies of the colon and rectum, are rare (1). Gastrointestinal Tract Duplications (GTD) account for about 2% to 4% of alimentary tract duplications and are most commonly observed in the abdominal cavity, involving distal ileum, stomach, duodenum, jejunum, colon, ileocecal, rectum, and even the appendix (2, 3). Most cases of GTDs are diagnosed in the first years of life by ultrasound and contrast medium, while severe cases may require Magnetic Resonance Imaging (MRI) or Computed Tomography (CT) scan (2, 3). The symptoms include acute abdominal symptoms, such as abdominal mass or distension, pain, vomiting, and weight loss in children (3) and most cases (about 75%) are cystic type while few are tubular (3).

Diagnosis of GTDs before childbirth can help prevent the need for emergency surgery and optimal management before the occurrence of complications, such as bleeding and obstruction (4, 5). On the other hand, patients with GTD may remain undiagnosed until adulthood, while the surgical management and diagnostic process may differ in adults (6).

Reports have indicated that less than 7% of all cases of GTDs occur in the colon (2) and tubular form of duplication is less frequent than the cystic form (3). In addition, the disease is very rare in adult patients (6). Therefore, the case presented here is one of the rare cases observed and reporting the patient's presentations, clinical course, procedure of diagnosis and treatment can help enrich knowledge in this regard.

Table 1. The Initial Laboratory Results of Serum Parameters of the Patient

Serum Parameter	Unit	Value	Normal Range
White blood cell	Mil/Cumm	8900	4500 - 11000
Red blood cell	Mil/Cumm	4.27	3.8 - 5.2
Hemoglobin	gr/dL	13.3	11.8 - 15.8
Platelets	gr/dL	303000	150000 - 450000
ESR	mm	42	2 - 20
Prothrombin time	Seconds	13.1	Control: 13.1
Partial thromboplastin time	Seconds	31.3	25 - 38
INR	-	1.00	1 - 1.3
Blood sugar	mg/dL	87	< 200
Blood urea nitrogen	mg/dL	11	8 - 20
Creatinine	mg/dL	0.6	0.5 - 0.9
Sodium (Na)	mEq/L	134	132 - 145
Potassium (K)	mEq/L	4.0	3.6 - 5.2
Chloride (Cl)	mEq/L	95	99 - 108
Bicarbonate	mEq/L	24	22 - 29

2. Case Presentation

The patient was a 31-year-old female, who was presented to our medical center (Pars Hospital, Tehran, Iran) with chronic abdominal pain and constipation from the childhood period. In her past medical history, she had a history of abortion about one month before, which required curettage, and a history of rhinoplasty. She mentioned no history of medication use.

In her physical examination, she had normal vital signs (blood pressure: 110/70 mmHg, pulse rate: 82/minutes, and respiratory rate: 20/minutes) and normal cardiac and pulmonary auscultation. In the initial laboratory examination, she had a normal cell blood count, coagulation tests, and blood chemistry, and a minor increase in Estimated Sedimentation Rate (ESR) (Table 1).

Colonoscopy was performed for the GI tract and the patient was referred for laparotomy by enterocolic fistula diagnosis. The patient was admitted to undergo laparotomy with suspicion of congenital colon anomaly. The day before surgery, she received 1000 cc of 1/3 to 2/3 Intravenous (IV) fluid three times per day and Ampicillin-sulbactam 3 gr IV. For the surgery, the patient received general anesthesia and was placed in lithotomic position and the abdomen was opened by a midline incision. During surgery, a 5 × 5 inflamed mass in the left colon attracted the surgeon's attention. Investigation of splenic flexure revealed an additional colon loop in posterior transverse and descending colon; half of the transverse colon was duplicated and the accessory lumen with a diameter of about 9 cm and length of 30 cm continued until the pelvis and had a blind end.

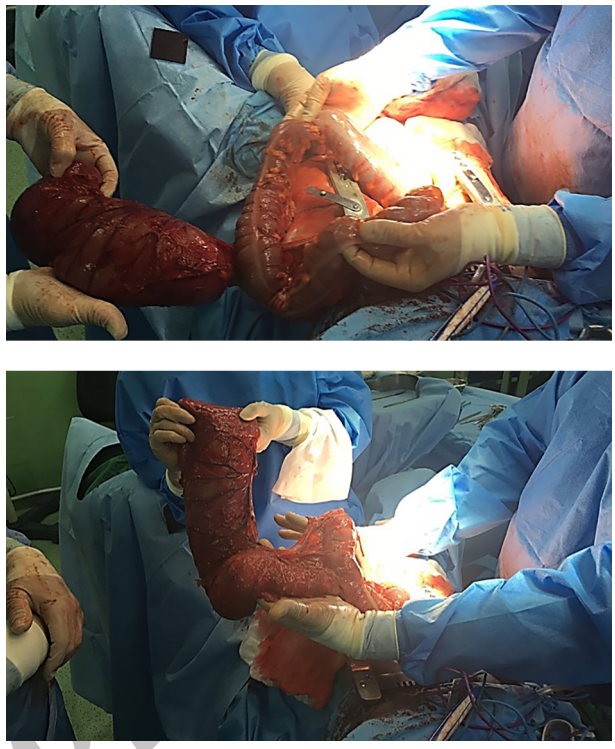

Figure 1. Surgical view of tubular duplication of colon with marked dilatation in the reported patient

The adhesions were released and the accessory loop was removed by segmental colectomy and close loop resection and sent for pathologic examination.

In a macroscopic evaluation, the specimen was a segmental colonic specimen (28 cm in length and 5 cm in diameter); the middle part was a distended blind intestinal loop, measuring 30 cm in length and 9 cm in diameter at the distal end (Figure 1). Mucosal continuity with the main specimen was noted, except for about 3 cm at the distal end, appearing ulcerated or fibrotic. The attached omentum measured 14 × 10 × 1 cm; five lymph nodes measuring 1.5 cm in greatest dimension were dissected. Representative sections were submitted in 14 blocks, from proximal and distal margins, random specimens, blind loop, and end of blind loop, lymph nodes, and omentum.

In microscopic examination, sections showed congested colonic wall with a focal area of mucosal ulceration, granulation tissue formation, fibrosis, and nonspecific chronic inflammation at the end of the blind loop without evidence of heterotopic mucosa or tumoral process. These results suggested a diagnosis of tubular duplication of the colon with marked dilatation, focal mucosal ulceration, granulation tissue, and mild chronic non-

specific inflammation. The included congested omentum showed no significant histopathologic changes. The final diagnosis of the patient was a congenital tubular duplication of the colon.

After discharge from the hospital, she had three times follow-up visits and all pre-operative problems were resolved without any complication.

3. Discussion

The present report describes the characteristics of a 31-year-old female, purely presenting chronic abdominal pain and constipation, finally diagnosed with complete duplication of the colon and successfully managed by segmental colectomy and close loop resection.

The advances in medical devices have enabled prenatal diagnosis of GTDs, while most of them remain asymptomatic until surgery (4, 5). The cases not diagnosed prenatally may reveal symptoms of obstruction or abdominal pain in early childhood (3) or remain undiagnosed until adulthood, found incidentally (6) or presented by symptoms (6-8). The symptoms may differ according to anatomic variances, such as site, size, presence of ectopic mucosa, inflammation, and adhesions/communications. Meanwhile, as most symptoms are non-specific, the diagnosis is not easy, especially in adults (9).

The most common site of GTDs is reported to be the ileum, while the colon consists of less than 7% of pediatric patients (3). Studying the frequency of manifestations in case reports of colonic duplication revealed different manifestations, as demonstrated in Table 2, including rectal bleeding or bloody stool (9), constipation (9, 10), and nausea/vomiting (11), while the patient presented here referred with the chief complaint of abdominal pain and constipation. The inflammation, dilatation, focal mucosal ulceration, and granulation tissue reported by microscopic examination in the patient showed the origin of her abdominal pain, besides the effect of compression of adjacent organs. Similar to the case presented here, Al-Jaroof et al. reported a case of a 33-year-old female with abdominal pain, without nausea/vomiting or chronic constipation, and unremarkable laboratory examination, for whom other diagnoses, including diverticulum, volvulus, and duplication cysts were considered as differential diagnoses, and final diagnosis was made by laparotomy (12). Consistent with this study, in the present report, the serum parameters were all within the normal range, and only an increased ESR was observed (Table 1) and the final diagnosis was made by laparotomy. The case reported by Sobhani et al. was an adult patient, who also had plain abdominal pain with normal serum parameters and unremarkable X-ray results (13). The final diagnosis in both the above-mentioned studies (12, 13), similar to the current report, was tubular colon duplication, diagnosed by exploratory laparotomy. These results suggest that abdominal pain could be the sole symptom of a colonic obstruction.

As far as the clinical symptoms of colonic obstruction are non-specific, physicians may request many investigations before surgery for accurate diagnosis, reporting different results (9-13, 15). Reports suggest that although CT scan could successfully confirm the diagnosis of a duplicated loop (Figure 2), there were indecisive results by colonoscopy, ultrasound, or X-ray (9, 10, 15). In the current case, the patient was diagnosed by performing colonoscopy as congenital anomaly of GI tract prior to surgery (enterocolic fistula). One of the reasons of misdiagnosis or unclear diagnosis by several imaging techniques could be the type of duplication, as the case presented here had a tubular type colonic duplication, while less than 25% of cases of colonic duplication are tubular and most cases (75% to 80%) are cystic (3). Most tubular colonic duplications are doubled (80%), as in the current case, while few can be Y-shaped (15), which may not be diagnosed by colonoscopy or simple X-ray. Also, as described, they may have variable anatomic and pathologic involvements, which can justify why some studies have reported CT and ultrasound as sufficient diagnostic tools (18), while others have reported them inappropriate (15).

The case presented here, similar to previous reports (12, 13), was diagnosed during surgery; the surgeon performed segmental colectomy and close loop resection in the current report. Most studies suggest complete resection of duplication with the relevant parts of the colon soon after diagnosis, to prevent future complications. Yet, as there are less than 100 cases reported in the world, the definite treatment approach is still controversial (20); some researchers suggest that colon duplication is a benign disease (10) and asymptomatic cases do not require radical surgical treatment (16), while other report risks of neoplastic change (19) or perforation (14, 17, 21). Although the patient referred at a late age, she had, fortunately, no evidence of heterotopic mucosa or tumoral process.

In conclusion, the report here explained that the diagnosis of GTDs should always be considered by physicians, in all parts of the abdomen, even colon, mainly a rare site for GTD, with even solely abdominal pain, and in all ages, including adulthood. Notably, positive family history is less likely to be observed in patients. Complete resection of duplication and the associated segments are suggested as the appropriate treatment for tubular colonic duplication.

Table 2. Characteristics of Case Reports Describing Adult Patients with Congenital Colonic Duplication

First Author	Patient's Age (Y)	Patient's Sex	Patient's Symptoms	Investigations	Type
Al-Jaroof (12)	33	Female	Right-sided colicky abdominal pain	X-ray, CT, MRI, barium enema	Tubular
Sobhani (13)	27	Male	Repeated episodes of abdominal pain in epigastric region	X-ray, CT	Tubular
Kyo (14)	20	Male	Intermittent right flank pain	Colonoscopy, CT	Both cystic and tubular
Banchini (15)	21	Male	Chronic constipation	Ultrasound, CT, small-bowel contrast study	Tubular
Kekez (10)	42	Female	Abdominal distension, chronic constipation	Colonoscopy, X-ray, anal manometry, CT	Tubular
Kim (16)	40	Female	Asymptomatic	Incidental finding on CT, confirmed by colonoscopy	Tubular
Kabay (11)	28	Male	Intermittent right flank pain, nausea and vomiting	Ultrasound, CT, renal tests	Tubular
Jung (17)	40	Female	Chronic pelvic pain and smelling vaginal discharge	Ultrasound, CT	Tubular
Fotiadis (9)	53	Male	Rectal bleeding, constipation	Colonoscopy, X-ray, CT	Cystic
Fotiadis (9)	45	Male	Fatigue, bloody stool	Radiography, colonoscopy	Cystic
Frittelli (18)	24	Male	Nausea and vomiting, abdominal pain	Ultrasound, CT	Cystic
Otomi (19)	26	Male	Upper left abdominal pain and fever	CT, gastrografin	-

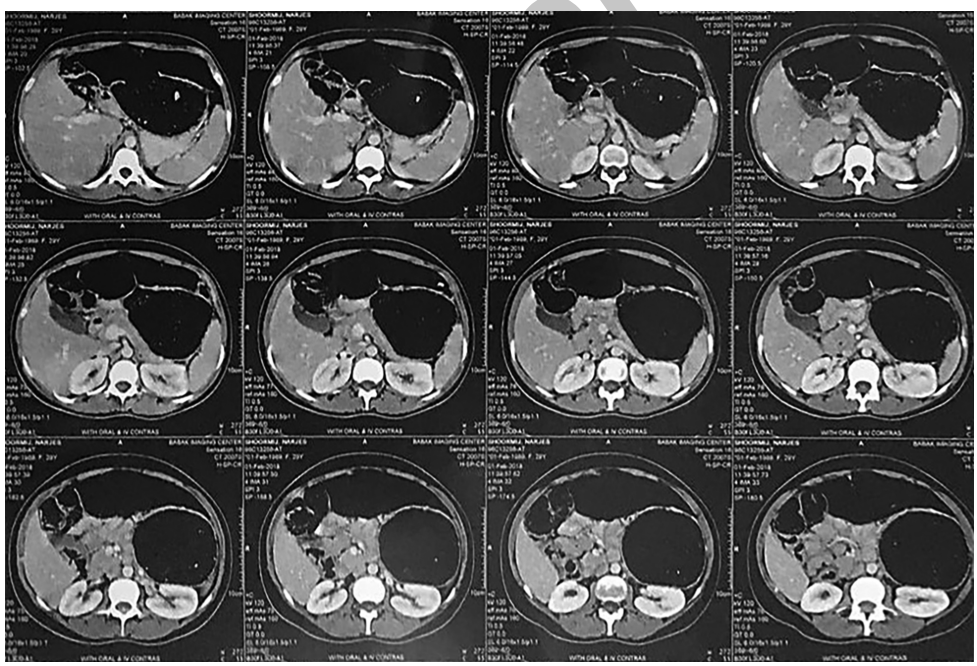

Figure 2. Magnetic Resonance Imaging (MRI) of the patient's abdomen

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