Sigmoid Duplication a Rare Cause of Colon Obstruction Due to Fecal Material Bezoar: A Case Report

Mahdi Alemrajabi [1], Morteza Khavaninzadeh [2], Mohammad Moradi [3*], Ali Dah Mardeh Ei [4]

[1] *Fellow of colorectal surgery. FCRDC. Iran University of Medical Sciences. Tehran. Iran.*
[2] *Hasheminejad Kidney Center (HKC), Iran University of Medical Sciences. Tehran, Iran*
[3] *Colorectal clinical fellow, Christie NHS Foundation Trust, Manchester, UK*
[4] *Department of general surgery, Shariati Hospital, TUMS, Iran*

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Abstract

Intestinal duplication is a rare congenital disorder that can occur anywhere in the gastrointestinal tract. The pathophysiology of duplications remains unclear, and various theories have been proposed. This report presents a case of a young man who came to the clinic with constipation and obstructive defecatory syndrome. Preoperative colonoscopy and imaging revealed no abnormalities in the colon. During the operation, a large colon mass was found and resected, and a primary colocolic anastomosis was performed laparoscopically. After specimen extraction, the colon was cut to assess the etiology. A congenital duplicated lumen of the sigmoid was found, with an accumulation of fecal material in the second lumen causing pressure and obstructing the main lumen. Duplications are a rare cause of intestinal obstruction and should be considered in the differential diagnosis of chronic or partial obstruction. This appears to be the first reported case of laparoscopic resection of sigmoid duplication in an adult man with chronic constipation in the literature.

Keywords: Colon Duplication, Constipation, Bowel Obstruction, Bezoar

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Introduction

Various congenital anomalies can affect the gastrointestinal (GI) tract. Most GI anomalies are detected early in life, and rare cases are not diagnosed until adulthood. In referral centers, 2-3 cases of GI duplications are reported annually [1]. Its incidence has been reported as 1 in 4500 live births and is detected in 0.2% of children [2]. The ileum and ileocecal valve are the most common sites, each accounting for 30%, followed by the jejunum (8%), colon (6%-7%), and rectum (5%) [3]. Duplications are sometimes associated with other malformations, which can facilitate diagnosis. However, some patients present with complications of duplications based on the involved part of the gut. These patients may experience chronic abdominal pain, constipation, a leading point for volvulus or intussusception, abdominal mass, or obstruction. Recently, it has been suggested that colon cancer may originate from such duplications.

Case presentation

The authors reported a case of a 40-year-old male who presented with chronic constipation. Despite receiving medical therapy for chronic constipation and pelvic floor biofeedback, there was no improvement. He had been suffering from chronic constipation for at least 10 years, with symptoms of straining, incomplete fecal evacuation, and pelvic fullness. Preoperative colonoscopy revealed only a mucosal prolapse of grade 2 to 3, which was confirmed by defecography. The patient was otherwise healthy and was scheduled for a laparoscopic ventral rectopexy.

At the start of the operation, a urethral duplication was detected when a Foley catheter was inserted. There were two meatuses. When the Foley catheter was inserted into the upper meatus, the tip of the catheter came out from the lower meatus. Hence, the Foley catheter was inserted into the lower meatus, which entered the bladder and urine was evacuated.

*** Corresponding author**: Dr. Mohammad Moradi
Colorectal clinical fellow, Christie NHS Foundation Trust, Manchester, UK
Email: mohammad.moradi@nhs.net

The operation was initiated with port insertion and insufflation to 14mmHg. During the operation, a large mass was noticed in the rectosigmoid junction. Therefore, a laparoscopic resection of the sigmoid was performed with a primary anastomosis due to suspicion of a colon tumor. After specimen extraction from a small Pfannenstiel incision, the specimen was cut. There was a duplicated lumen filled with fecal material bezoar and a compressive effect on the main lumen causing partial obstruction. The second lumen was separated from the main one by a true colonic wall. Pathologic examination of the specimen confirmed a muscular layer between the two mucosal layers. Written informed consent was obtained and the patient was assured regarding the anonymity of data. After one year of follow-up, the patient had no constipation or any complaints. The authors believe the patient's symptoms were attributed more to the bezoar and partial obstruction than to an internal grade 2-3 mucosal prolapse.

Discussion

There are very few cases of GI duplication presented with constipation in the literature, similar to the authors' patient. Kekez et al. reported a 42-year-old patient with chronic constipation and Hashimoto thyroiditis. No reason for chronic constipation had been found, and constipation was attributed to his underlying hypothyroidism. He had undergone a complete workup over eight years without any improvement in his symptoms. Finally, an abdominal CT scan following abdominal distention revealed a colonic pathology, but the definitive diagnosis of duplication of the transversal colon was made intraoperatively [4]. This was similar to the authors'

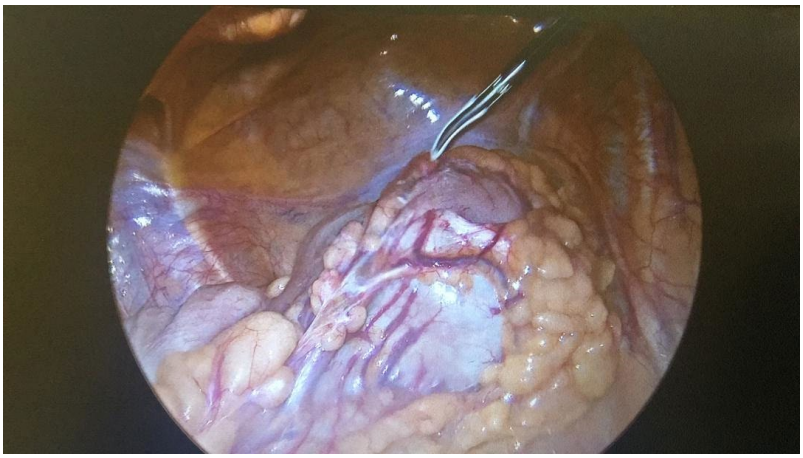

Fig. 1. Laparoscopic View of Sigmoid Showing a Mass Like Lesion

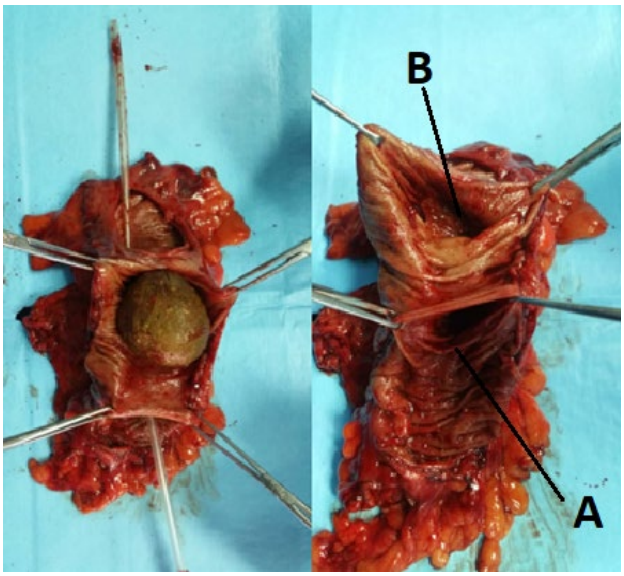

Fig. 2. Duplicated Lumen of Sigmoid After Specimen Extraction (A indicates the true lumen and B the duplication)

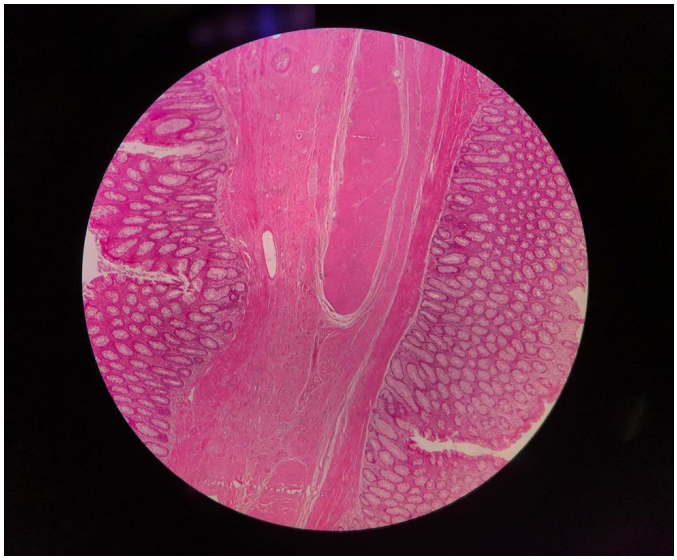

Fig. 3. Pathologic Examination of the Specimen Confirming a Muscular Layer Between the Two Mucosal Layers.

case in terms of age and primary symptoms, who remained undiagnosed despite several diagnostic workups.

In 2018, Xin Wu described a 25-year-old woman who presented with abdominal pain, and a tubular colonic duplication was found intraoperatively [5]. Kyo et al. in 2016 presented a case of ascending colon duplication in a 20-year-old male that caused intussusception. They performed a laparoscopic resection without any complications [6]. Additionally, a case series reported two cases of colonic duplication in adults who presented with rectal bleeding on admission. They concluded that duplications must be considered in the differential diagnosis of lower GI bleeding in adults [7].

To the authors' knowledge, no report exists regarding colon duplication incidentally found during laparoscopic rectopexy in a patient with chronic constipation. Anatomically, this duplicated lumen may or may not communicate with the normal lumen. The dividing wall between the two lumens may be muscular or simply a double layer of epithelium. However, as seen in the authors' patient, there was a true wall between the two lumens.

Gross et al. [8] described four variations in the shape of duplications, including a tubular structure, a double-barreled structure, a cystic structure lying in the peritoneal cavity attached by a mesenteric stalk, and a spherical lesion. The most common type is the cystic one, constituting 90%-95% of patients. There is a classification for such anomalies named as McPherson, which includes type I (simple cysts), type II (diverticula), and the most common type III (tubular colonic duplication) [9].

The exact pathophysiology of colon duplication is not yet known. However, some factors such as trauma or hypoxia have been proposed [10]. Moreover, the coexistence of other malformations such as vertebral or genitourinary ones may explain spinal dysraphism as the underlying congenital etiology [11]. Some others may be associated with a pancreatic cyst [12].

The diagnosis in childhood is usually made due to abdominal pain, constipation, a leading point for intussusception, lower GI bleeding, or a palpable abdominal mass. However, the diagnosis in adults is challenging. In the authors' case, fecal material accumulation in the second lumen causing partial obstruction and chronic constipation, and a mass-like lesion in laparoscopic view helped to secure the diagnosis. Duplications are rarely diagnosed above 2 years of age.

Radiographic imaging such as ultrasonography, CT scan, or colonoscopy performed for other reasons may detect the malformation. However, preoperative colonoscopy in the authors' patient found no abnormality in the colon. Despite this, a normal colonoscopy cannot rule out the diagnosis as the scope might only pass through the true lumen. Therefore, a colonic duplication must be kept in mind despite a normal preoperative diagnostic assessment. The authors believe a contrast enema study might help to detect GI duplications, particularly in those where there is a communication between a duplication and a true lumen.

It has been recommended to resect colonic duplication if diagnosed due to a small risk of malignancy. Despite this, there is no clinical trial or a long-term cohort to prove the higher risk of malignancy, but it should be kept in mind. Colonic

resection could be performed by a laparoscopic approach easily. As seen in the authors' case, colon duplication can be missed until higher ages even despite a concurrent anatomical abnormality. The patient had a duplicated urethra but did not seek urologic consultation.

Conclusion

In conclusion, despite its rarity, colonic duplication should be considered in the differential diagnosis of constipation or abdominal mass, especially when there is a concurrent anatomic abnormality in the vertebra or genitourinary tract.

Conflict of Interest

The authors declare no conflict of interest.

Funding Statement

None.

Ethical Approval

We obtained a local approval from our institute to report the case.

Consent

An informed written consent was obtained from the patient. His data remained confidential and anonymous.

Guarantor

Mohammad Moradi the corresponding author would be the guarantor of the content to be published.

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