

CASE REPORT

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Multiple Small Bowel Diverticula Were an Unexpected Finding During Laparoscopic Enterectomy for Crohn's Disease

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ABSTRACT

Introduction: Small bowel diverticulosis (SBD) is a rare entity. Although it is usually an asymptomatic condition, clinical manifestations may vary from non-specific clinical signs to severe and complicated disease. The coexistence of SBD and Crohn's disease (CD) is rarely reported in the current literature. **Aim:** We present a rare case of concomitant Crohn's disease (CD) and SBD in a male patient, where multiple jejunal diverticula were an incidental intraoperative finding. Preoperative evaluation with magnetic resonance enterography (MRE) failed to recognize the coexistence of these two entities. Surgeons should be aware of the possibility of this rare situation. **Case report:** A 52-year-old Caucasian male diagnosed with CD was referred to our department for surgical intervention due to an ileal stricture. The patient reported no past medical history, except for a few episodes of bloody diarrhoea during a three-year period. The index colonoscopy revealed luminal narrowing in the ileum at approximately 70 cm proximal to the ileocaecal valve, and biopsies revealed findings compatible with CD. Clinical examination and laboratory tests were unremarkable one day before surgery. The patient underwent laparoscopic segmental resection of the affected part of the ileum. Intraoperatively, multiple non-inflamed diverticula along the jejunum extending from the Treitz ligament to the proximal ileum were recognized. Our patient had an uncomplicated post-operative course and was discharged on the fifth post-operative day. Pathological examination revealed features compatible with CD in the active phase. The patient was referred to his gastroenterological team for further consultation regarding the appropriate post-operative management. **Conclusion:** Concomitant CD and SBD is a rare condition, and the differential diagnosis may be challenging due to overlapping symptoms.

Keywords: Crohn's disease, case report, magnetic resonance enterography, small intestine diverticulosis

1. INTRODUCTION

Diverticulosis of the jejunum is a rare clinical condition, usually presenting in elderly patients, whereas males seem to be affected more frequently than females (1). A small intestine diverticulum is a false diverticulum formed from an outpouching of the mucosa in the antimesenteric border of the small bowel following the pathway of the visceral vessels (2).

Although they are usually asymptomatic, clinical manifestations include gastrointestinal haemorrhage, pain, obstruction and peritonitis in an acute presentation or pain, nausea and malnutrition as a chronic form of the disease (3). Multiple small bowel diverticula (SBD) is less common in comparison with colonic diverticula, and the incidence rate is reported at

2.0-2.3%; SBD are mainly diagnosed as an incidental finding during enteroclysis (4).

The coexistence of multiple SBD and Crohn's disease (CD) is a rare condition (5, 6). We report the case of a 52-year-old male who underwent laparoscopic enterectomy in our surgical department due to stricturing CD of the terminal ileum with multiple jejunal diverticula revealed intraoperatively.

2. AIM

Small bowel diverticulosis (SBD) is an acquired condition and usually has an asymptomatic course, although it may present with non-specific symptoms or severe diverticular complications. We present a rare case of concomitant Crohn's disease (CD) and SBD in a male patient,

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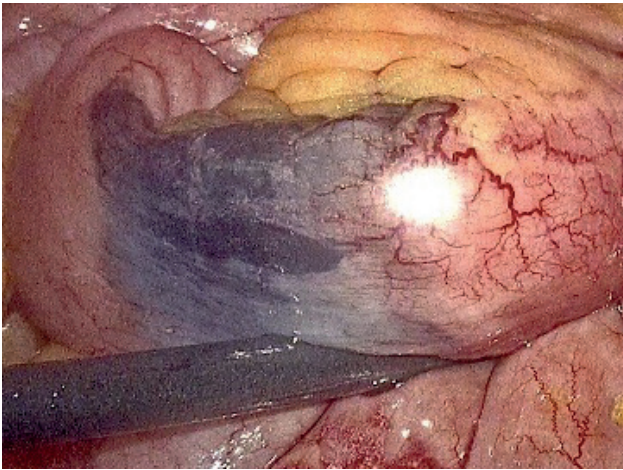


Figure 1. Intraoperative photograph showing the terminal ileum marked with blue dye.

mal white blood cell count of 8.8×10^3 cells/ μ L, a platelet count of 216×10^3 cells/ μ L, a normal CRP level of 0.1 mg/dl and an albumin level of 4.6 g/dl.

The extent of disease was preoperatively assessed with ileo-colonoscopy and magnetic resonance enterography (MRE). The aforementioned stenotic region in the ileum at approximately 70 cm proximal to the ileocaecal valve was present during ileo-colonoscopy; there was an additional finding of a large ulcer in the terminal ileum, which was marked with blue dye (Figure 1). The biopsies showed histologic features of mild chronic ileitis without indices of dysplasia or malignancy. The MRE revealed a long ileal segment demonstrating asymmetric mural thickening for a length of approximately 10 cm, with a homogenous enhancement pattern and imaging findings in keeping with possible pseudosacculations/pseudodiverticula formation and without prestenotic dilatation (Figure 2).

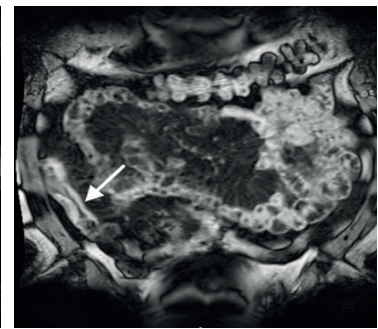
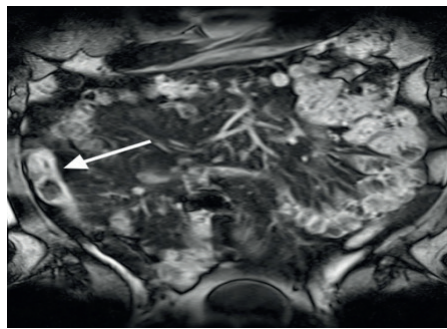
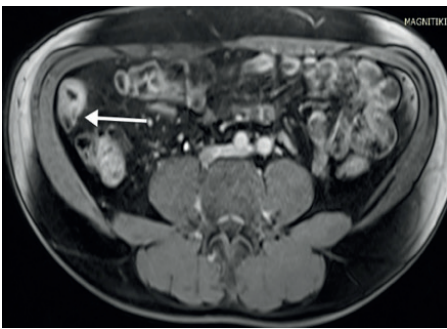


Figure 2 (a, b, c). Magnetic resonance enterography images, coronal (A, C) and axial (B) plane; T1 weighted 2D FLASH fat saturated images after gadolinium administration demonstrating homogenous enhancement of the asymmetrically thickened long ileal segment with the presence of pseudosacculations (A, B).

where multiple jejunal diverticula were an incidental intraoperative finding. Preoperative evaluation with magnetic resonance enterography (MRE) failed to recognize the coexistence of these two entities. Surgeons should be aware of the possibility of this rare situation.

3. CASE REPORT

A 52-year-old Caucasian male was referred to our department for surgical intervention due to ileal stricture as a complication of CD. The patient reported no past medical history, but he could recall a few episodes of bloody diarrhoea during the three-year period before the CD diagnosis was established; diagnostic evaluation with colonoscopy was negative for gastrointestinal tract abnormalities at that time. He had shown poor compliance with the proposed medical treatment, and he had not received any kind of medication since the onset of CD. The index colonoscopy, at which CD was diagnosed, revealed an area of luminal narrowing in the ileum at approximately 70 cm proximally to the ileocaecal valve. Biopsies were taken, and the pathologic report was indicative of CD.

On admission, one day before surgery, the patient was afebrile, with regular heart and respiratory rates. During abdominal examination, he had regular bowel sounds with no tenderness or other signs of peritoneal inflammation, whereas the digital rectal examination did not reveal any pathological findings. Laboratory tests revealed a normal haemoglobin level of 14.6 g/dl, a nor-

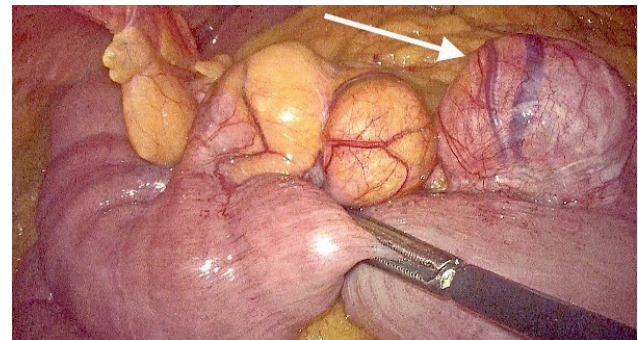


Figure 3. Intraoperative photograph showing non-inflamed diverticula along the jejunum.

The patient underwent three-dimensional laparoscopy (3D HD Storz system, Germany), where segmental resection of the affected part of the ileum (approximately 30 cm) was performed with end-to-end double-layer ileo-ileal anastomosis. During careful inspection of the small and large intestine for lesions compatible with CD, we found multiple non-inflamed diverticula along the jejunum extending from the Treitz ligament to the proximal ileum. (Figure 3, Video 1). Our patient had an uncomplicated post-operative period and was discharged on the fifth post-operative day.

Histopathological examination of the specimen showed focal inflammation and focal crypt architectural distortion compatible with CD in the active phase. The final diagnosis of the presented case was the stricturing phenotype of CD in the terminal ileum (based on the

Montreal Classification of CD) with concomitant multiple SBD. The patient was referred to his gastroenterological team for further consultation regarding the appropriate post-operative management. At the 10-month follow-up, our patient was asymptomatic and without new episodes of intestinal haemorrhage.

4. DISCUSSION

Jejunal diverticulosis has been a well-known condition for approximately three hundred years; the first descriptions were made by Somerling in 1794 and Sir Astley Cooper in 1807. Understanding the differences in epidemiology, aetiology and behaviour between diverticular disease of the small intestine and the colon plays an important role in decision making regarding the proper management (7).

First, jejunal diverticulosis is a rare condition, and the incidence rate of the disease is reported at approximately 2.0%-2.3% (4, 8). More recent literature data are inconsistent regarding the sex distribution; elderly patients and female patients seem to be at higher risk for developing SBD. However, it should be kept in mind that SBD can affect individuals at any age (3, 9). Second, the behaviour of the disease has differences in comparison with colonic diverticula regarding the presentation, work-up and treatment. Clinical manifestations can vary from nonspecific chronic pain with malabsorption to life-threatening complications such as diverticulitis with perforation, intestinal obstruction and haemorrhage. Furthermore, patients with multiple SBD are in greater danger of developing life-threatening complications; however, extended surgical resections should be avoided due to the danger of short bowel syndrome, and enterectomy should only include the bowel loop with the complicated diverticulum (10, 11).

In a review article, Makris et al. described the difficulty of clarifying the exact source of haemorrhage in previously asymptomatic patients when the source is a diverticulum of the small intestine (3). We can speculate that clinical suspicion for SBD as the source of recurrent intestinal haemorrhage in a patient with inflammatory bowel disease is extremely low. MREs performed during the disease course did not reveal the existence of multiple SBD, whereas endoscopic findings in the terminal ileum were suggestive of ulceration as the obvious source of the haemorrhage. Multiple diverticula in the mesenteric border of the jejunum were an incidental finding during laparoscopy.

Searching the literature, we found many original articles and case reports reporting multiple SBD, but data regarding the coexistence of inflammatory bowel disease and SBD were scarce. The first reports, including patients with simultaneous Crohn's disease and small intestine diverticulosis, were published in 1938 from Barrington-Ward and Norrish and in 1958 from Ekman with different assumptions regarding the nature of the disease (12, 13). Subsequent articles suggesting an overlap between SBD and Crohn's disease were published. In their recent review, Paeschke et al. described the inflammatory process in the enteric wall as a possible substrate

for diverticula formation (14). Gledhill et al. presented 11 patients with diverticulosis and inflammatory bowel disease in 1998 and tried to show the connection in pathological findings between a "Crohn's-type response" and diverticulosis. Only two patients had clinical manifestations suggestive of Crohn's disease, whereas the others had manifestations suggestive of diverticular disease. The authors suggested that more accurate clinical and radiological diagnostic criteria must be considered in order to discriminate between the two pathological entities (15). Shepherd, in 1996, suggested that in complicated diverticular disease, a Crohn's-type response is present as mimicry (16).

Consequently, the possible association between CD and SBD was examined in terms of identifying common genetic predisposition for both diseases. This was partly evaluated in the article by Connelly et al., who proposed that similar but distinct genetic predispositions are shared between CD and diverticulosis; overlapping haplotypes were found in the *tumour necrosis superfamily 15* gene [*TNFSF15*] (17). To our knowledge, there are no other studies searching for common genetic backgrounds between the two entities; therefore, we believe that further research is needed to investigate the possible associations between CD and SBD.

5. CONCLUSION

In conclusion, we believe that, despite the rarity of jejunal diverticular disease, physicians should keep in mind that overlapping clinical manifestations between CD and other pathological entities may occur. An endoscopic and histological diagnosis of CD should always be accompanied by thorough gastrointestinal tract mapping via radiological modalities in patients who are candidates for surgery in order to choose the best treatment option and avoid unexpected findings intraoperatively.

- **Declaration of patient consent:** Informed written consent was obtained from the patient for publication of this report and any accompanying images and videos.
- **Author's contribution:** Sotirova I and Papaconstantinou I were the patient's surgeons, reviewed the literature and contributed to manuscript drafting; Papalouka D reviewed the literature and contributed to manuscript drafting; Gourtsoyanni S analysed and interpreted the imaging findings and contributed to manuscript drafting; Christodoulou D was the patient's consultant gastroenterologist, reviewed the literature and drafted the manuscript; Gklavas A was responsible for the revision of the manuscript for important intellectual content; all authors issued final approval for the version to be submitted.
- **Conflict of interest statement:** The authors declare that they have no conflict of interest.
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