Colonic Diaphragm in a Patient with Rheumatoid Arthritis – A Case Report from the Caribbean
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ABSTRACT

Colonic diaphragm is a rare complication of chronic use of non-steroidal anti-inflammatory drugs. Though benign, life-threatening complications such as intestinal obstruction and anaemia may ensue. Distinguishing colonic diaphragm from other causes of intestinal obstruction such as malignancy is often difficult, with patients often requiring surgical intervention to establish the diagnosis. We present a case of colonic diaphragm with a short review of the clinical and pathologic features.

Keywords: Colonic, diaphragm, NSAID, obstruction

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West Indian Med J DOI: 10.7727/wimj.2015.488
INTRODUCTION

Colonic diaphragm disease is a rare complication of the chronic use of non-steroidal anti-inflammatory drugs (NSAID). NSAID are among the most widely used medications in the world with more than 30 million people worldwide using them on a daily basis (1). Upper gastrointestinal complications of chronic NSAID use are well established, occurring in as much as 25% of patients. However, colonic ulceration and diaphragm disease has also become recognized as a major adverse event associated with NSAID use (2,3). Colonic diaphragms, though benign, can have serious patient implications such as intestinal obstruction and anaemia (3). Depending on the patient’s presentation, management may be through endoscopic or surgical means. Surgical intervention may be required in cases of colonic obstruction or for suspected malignant disease. The first case report and discussion of a patient with colonic diaphragm disease from the Caribbean is presented.

CASE REPORT

A 55-year-old woman known to have rheumatoid arthritis being managed with chronic NSAID therapy (diclofenac) for eight years presented with a three-week history of constipation, abdominal distension and cramping abdominal pain. This was associated with intermittent vomiting. She had experienced an indeterminate amount of weight loss over the preceding three years. There was no record of fever or per rectal bleeding. Her past surgical history included total abdominal hysterectomy and bilateral hip replacements.

Physical examination revealed anaemia, mild dehydration and arthritic changes to both hands. Vital signs were normal. The abdomen was mildly distended, but non-tender with normal bowel sounds and a normal digital rectal examination.
The clinical assessment was that of uncomplicated partial intestinal obstruction possibly due to malignancy, benign stricture or adhesions. Blood investigations confirmed anaemia (hemoglobin 10mg/dl). Other laboratory tests were normal. Abdominal radiographs showed dilated loops of large and small bowel and an obstructing rectosigmoid lesion was identified on computed tomography scan of the abdomen. There was no evidence of liver or peritoneal metastases. A flexible sigmoidoscopy was performed, which confirmed the presence of a stricture at approximately 30cm from anal verge. The sigmoidoscope was unable to pass beyond the lesion.

At laparotomy, the lesion was identified and bearing in mind the possibility of malignancy, an oncologic left hemicolectomy was performed. A decision was made not to re-establish intestinal continuity due to her poor nutritional status and chronic steroid use. An end colostomy was fashioned with closure of the rectal stump. Her post-operative period was complicated by a non-ST segment elevation myocardial infarction day two post-procedure. This was successfully managed without any sequelae. She was discharged home one week post-surgery with a functional colostomy after tolerating normal diet.

Gross pathological examination revealed a left hemicolectomy specimen, which exhibited a 7.0 x 4.0 x 5.0 cm area of serosal thickening along the mesenteric border of the sigmoid colon. On opening, this thickened area corresponded to a valve-like band of mucosal thickening (Figure 1), traversing the lumen with proximal dilatation of the colon and associated inspissated faecoliths. Numerous wide-mouthed diverticula were also present throughout the colon. Histologically, the valve-like band revealed dense submucosal fibrosis and mild chronic inflammation typical of colonic diaphragm (Figure 2). Additionally, there was evidence of diverticulitis with paracolic abscess formation. There was no evidence of malignancy.
DISCUSSION

Diaphragm disease is a term used to describe the appearance of thin valve-like strictures within the bowel (4). It has been described more often occurring in the small intestine but it rarely occurs in the large bowel (5). Less than 50 cases of colonic diaphragm have been reported in the literature (6). Colonic diaphragm is a rare but benign disease, the natural history of which has not been fully elucidated. It is thought, however, to be the end-point of a spectrum of manifestations of colonic enteropathy, attributable predominantly to chronic NSAID use (7). Debenham described the first case of NSAID-induced colonic damage in 1966 (8) while colonic diaphragm disease was first described in 1989 by Sheers and Williams (9). Colonic diaphragms have also been described in children with cystic fibrosis treated with pancreatic enzyme supplements coated with methacrylic acid copolymer (10) as well as patients taking potassium supplementation. Colonic strictures have also been reported by Keating et al in 2 patients with vasculitis secondary to systemic lupus erythematosus and rheumatoid arthritis (11).

Similar to the findings noted in the present case, the gross pathology is one of a shelf-like or septal-like lesion within the lumen of the bowel, which may be complete or partial (12). The strictures can be difficult to differentiate macroscopically from cancer, diverticular disease and Crohn’s disease (13). Microscopically, there is a characteristic histological appearance of submucosal fibrosis, chaotic arrangement of smooth muscle fibers, and vascular and neural elements resembling hamartomas (6). There is no mucosal hyperplasia or dysplasia.

The pathogenesis of colonic diaphragm disease is poorly understood. Theories proposed attribute the changes to the local effects of chronic exposure to NSAID, resulting in inflammation, ulceration, healing with fibrosis and stenosis and consequent diaphragm formation (14). Clustering of the lesions at Bauhin’s valve in the cecum and ascending colon supports the hypothesis that the lesions
are not caused by a systemic process, but rather by a local injurious effect of the NSAID (15). A theory for the enterocyte damage proposes that there is uncoupling of oxidative phosphorylation in mucosal mitochondria, which causes depletion in the stores of adenosine triphosphate. This leads to increased mucosal permeability and efflux of calcium ions. The damaged enterocytes are also susceptible to further injury by bile acids, enzymes and bacteria because they are prevented from converting arachidonic acid into prostaglandins due to inhibition of cyclooxygenase activity (16). Mucosal invasion by bacteria releases chemotactic factors that attract neutrophils into the mucosa and submucosa leading to ulceration and strictures (15). A small case series suggested that the effects of the NSAID are not likely to be dose related because of the variation in doses that affected patients were treated with. However, there seems to be a temporal relationship between the duration of NSAID use and the manifestations in the colon (17). Our patient was being treated with NSAIDS for eight years.

The highest incidence is in the seventh decade of life with a female preponderance (18). Patients may present with only a history of chronic microcytic anaemia secondary to low grade bleed and iron deficiency or with symptoms of partial or complete intestinal obstruction coupled with a history of chronic NSAID use for an average of five years. These findings were congruous with our patient except for a younger age of presentation. Diclofenac has been identified as the most commonly implicated NSAID in colonic diaphragm disease (18).

As in the present case, based on the clinical presentation and difficulty in distinguishing colonic diaphragm from malignancy, surgical intervention is often required. Other presenting symptoms or signs of colonic diaphragm disease include change of bowel habit, weight loss, and gastrointestinal bleeding (1). Diagnosis can be made based on the history and colonoscopy findings. The physician must be aware of the condition, however, and have a high index of suspicion to avoid
misdiagnosis because of its similar presentation to diverticular stricture, inflammatory bowel disease and colon cancer.

Management depends on the presentation of the patient. In an emergency presentation of complete obstruction, resection and anastomosis is the preferred option. However, in partial obstruction or in asymptomatic cases where the cause is identified during colonoscopy, and biopsy has confirmed benign disease, endoscopic balloon dilatation has been shown to be successful (18). The morbidity and mortality associated with surgery can be high especially since this cohort of patients is usually malnourished, chronically ill and may have been on long-term steroids. As such, balloon dilatation is a favorable alternative (19). This method has been shown to be effective and has a low complication rate (20). To prevent further disease progression or recurrence it is also necessary to discontinue NSAID use (6). The non-specific colitis and colonic ulceration resolve on withdrawal of the NSAID therapy. However, the formation of strictures or diaphragms is irreversible and a definitive procedure is required (1). This patient may have been amenable to endoscopic balloon dilatation if colonic diaphragm was considered in the differential diagnosis. At the time, this was not suspected and therefore segmental resection with fecal diversion was performed.

CONCLUSION
We report the first case of colonic diaphragm recognized in the English-speaking Caribbean. Colonic diaphragmatic disease, though a benign condition, may have serious surgical implications. With the wide availability and frequent use of NSAID, the low prevalence of this condition suggests that it may be under-diagnosed. Increasing awareness of this clinical phenomenon enables early endoscopic diagnosis to be established and avoid confusion with other conditions such as inflammatory bowel
disease or colon cancer. This would lead to the appropriate management and cessation of the offending agent without subjecting the patient to unnecessary surgical intervention and possible complications.
REFERENCES


Figure 1. Gross image of left hemicolectomy specimen bivalved to demonstrate the colonic diaphragm.

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Figure 2 (a). Low power view of adjacent normal mucosa and submucosa.
Figure 2 (b). Low power view of diaphragm with submucosal fibrosis and chronic inflammation

Figure 2 (c). Low power view showing increased vascularity and fibrosis.