# Até onde devemos insistir no desenrolamento e descompressão endoscópicos num volvo recorrente da sigmóide?

How far should we go with endoscopic detorsion and decompression in a recurrent sigmoid volvulus?

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# RESUMO

Introdução: Após descompressão endoscópica, o volvo do cólon recorre em 75% dos casos. Cada recidiva acarreta um risco acrescido de complicações e morte.

**Objetivo:** Sensibilizar para os riscos de protelar o tratamento cirúrgico definitivo de volvo cólico recorrente.

Resumo: Mulher, 51 anos, admitida por distensão e dor abdominal. Antecedentes de megacólon idiopático, com volvo recorrente do cólon sigmóide, submetido a descompressão endoscópica. A radiografia do abdómen mostrou ansa de sigmóide dilatada, com sinal de "grão de café". A sigmoidoscopia confirmou o volvo, apresentando sinais de necrose entre os pontos de torção. A laparotomia exploradora revelou distensão cólica marcada, com gangrena da ansa torcida, optando-se pela colectomia total com ileostomia terminal.

**Conclusão:** Uma estratégia cirúrgica profilática não-emergente em doentes com volvo recorrente deve ser considerada para prevenir complicações graves e cirurgia emergente com elevada morbimortalidade.

# INTRODUCTION

Large-bowel obstruction in adults is mostly caused by colorectal cancer, diverticular disease and colonic volvulus.<sup>1,2</sup> In western world, colonic volvulus accounts for up

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### ABSTRACT

*Introduction:* After endoscopic decompression, colonic volvulus recurs in 75% of cases. Each relapse carries an increased risk of complications and death.

**Objective:** To highlight the risks of delaying definitive surgical treatment for recurrent colonic volvulus.

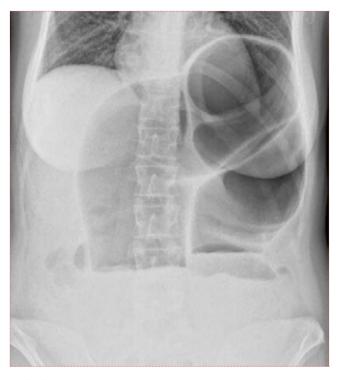
**Clinical case:** A 51-year-old woman was admitted for abdominal pain and distension. She had history of idiopathic megacolon, with recurrent sigmoid volvulus, submitted to endoscopic decompressions. The abdominal X-ray showed a dilated sigmoid loop, with a "coffee bean" sign. Sigmoidoscopy confirmed volvulus, with signs of necrosis between the points of torsion. Surgical exploration revealed marked colonic distension with gangrene of the twisted loop; total colectomy with end-ileostomy was performed.

**Conclusion:** A non-emergent prophylactic surgical strategy in patients with recurrent volvulus should be considered to prevent serious complications and emergent surgery with high morbimortality.

# to 3.5% of all cases of bowel obstruction.<sup>3,4</sup>

Sigmoid volvulus is the most common type of intestinal volvulus.<sup>5</sup> It is caused by the torsion of the sigmoid loop around its mesenteric axis, resulting in a closed-loop obstruction.<sup>2</sup> Without a prompt reduction, the closedloop obstruction results in an increase of intra-luminal pressure with compromise of the blood supply of the twisted segment, leading to ischemia, gangrene and perforation, carrying a significant risk of morbidity and mortality.<sup>6,7,8</sup>

The management of colonic volvulus includes assessment of the viability of the twisted colon segment, relief of the colon obstruction, and prevention of recurrent attacks. The most recent published guidelines<sup>1,3</sup> recommend non-surgical detorsion and decompression with sigmoidoscopy (with or without placement of a



**FIGURE 1**. Abdominal X-ray showing a distended loop of sigmoid colon, resembling a "coffee bean".

decompression tube) as the first therapeutic option in the management of uncomplicated sigmoid volvulus. On the other hand, emergency surgical treatment at the time of acute presentation is the appropriate treatment in patients with sigmoid volvulus associated with gangrene, perforation, or unsuccessful endoscopic reduction.

The success rate of endoscopic detorsion in sigmoid volvulus can reach 95%.<sup>2,4,7,8</sup> However, without a subsequent definitive surgical treatment, colonic volvulus tends to recur. Recurrent volvulus was described in approximately 25-75% of patients after a successful endoscopic detorsion.<sup>1,2,5-9</sup> Thus, the current guidelines advocate sigmoid colectomy after the resolution of the acute phase of sigmoid volvulus.<sup>1</sup>

We present a case of a recurrent sigmoid volvulus in a patient with a major risk factor for recurrence – megacolon. The purpose of this case report is to increase the awareness towards a prophylactic surgical strategy in patients with recurrent sigmoid volvulus in order to prevent potential life-threatening complications, such as bowel ischemia and perforation, and to avoid the unfavorable outcomes associated with an emergency surgery.

### **CASE PRESENTATION**

A 51-year-old Caucasian woman, born and residing in Portugal, was admitted at the emergency department complaining of nausea, abdominal cramping pain, distension, and constipation during the past 5 days.

The patient had a history of chronic long-standing constipation and an idiopathic megacolon diagnosed in 2012, complicated with multiple episodes of recurrent sigmoid volvulus. She presented at the emergency department at least nine times over the past 8 years with symptoms and signs compatible with sigmoid volvulus. At all times, the sigmoid volvulus was managed with endoscopic detorsion and decompression.

Initial physical examination revealed a markedly distended and tympanic abdomen, but only slightly tender on palpation, without signs of peritoneal irritation. Bowel sounds were decreased. Digital rectal examination revealed an empty rectum. The patient was afebrile and hemodynamically stable. Laboratory data on admission did not show any abnormalities other than leukocytosis (15 x 10<sup>9</sup>/L). Plain abdominal X-ray showed a dilated sigmoid loop with the classic "coffee bean" sign and lack of air in the rectum (Figure 1), suggestive of sigmoid volvulus. There were no signs of free intraperitoneal air.

The patient underwent a flexible sigmoidoscopy, with minimal CO<sub>2</sub> insufflation, which revealed an abruptly spiral convergence of the colonic folds ending in a pinpoint occlusion ("whirl" sign), at the level of the recto-sigmoid transition (Figure 2). After the endoscope passed with some resistance through the point of torsion, an overly dilated colon was seen. The mucosa between the distal and proximal points of torsion exhibited a violaceous color with dark spots and blood suggesting necrosis (Figure 3). After the endoscopic intervention, a significant improvement in abdominal distension was noticed. A decompression tube was not left in place.

Six hours after admission, the patient presented with a significant worsening of abdominal pain, with generalized tenderness on palpation, but without guarding or rebound tenderness. At this time, she presented with hypotension, tachycardia and metabolic acidosis on blood gas analysis. The patient had a favorable response to fluid challenge, not requiring vasopressors.



**FIGURE 2**. Endoscopic view of the typical appearance of the twisted colon mucosa in a sigmoid volvulus ("whirl" sign).



**FIGURE 4**. Macroscopic appearance of the megacolon in the operating room.

Giving the endoscopic findings and the deterioration of clinical condition, the patient was submitted to an emergent surgical exploration that revealed a purulent peritoneal fluid in abdominal cavity and a marked colonic distension, especially in the cecum and sigmoid colon, with gangrene of the twisted sigmoid loop but without any sign of mural defect suggestive of perforation (Figure 4). Taking into account these intraoperative findings, and considering the medical history of recurrent sigmoid volvulus and the concomitant megacolon, the patient was submitted to a total colectomy with an end-ileostomy (giving the high risk of anastomotic dehiscence).

Subsequent histological examination revealed a trans-



**FIGURE 3**. Endoscopic view of obstruction site with signs of ischemia of the twisted colon mucosa.

mural ischemic necrosis of the transverse, descending and sigmoid colon, with no reference to aganglionic lesions.

The patient had an uneventful postoperative course and was discharged on postoperative day 10. Six months later she underwent a successful intestinal reconstruction with an ileorectal anastomosis. She remains clinically asymptomatic over one year of follow-up.

### DISCUSSION

The pathogenic mechanisms of sigmoid volvulus are yet not fully understood.<sup>6</sup> Numerous predisposing factors have been described, including anatomic factors, chronic constipation, high-fiber diet, frequent laxative use, neurological disorders, colonic dysmotility, and adhesions from previous abdominal surgery.<sup>34,6</sup>

An anatomical predisposing factor, dolichosigmoid, which is a redundant sigmoid colon with a long and narrow mesenteric attachment, is the most important factor in the development of both primary and recurrent sigmoid volvulus.<sup>9</sup> This condition may be congenital, acquired, or a combination of the two mechanisms.<sup>5</sup> Chronic constipation, with chronic fecal overloading, may induce progressive elongation and dilatation of the sigmoid colon, resulting in a long redundant loop, contributing to sigmoid torsion.<sup>10</sup>

Since sigmoid volvulus is also associated with other numerous medical and neuropsychiatric comorbidities, it is believed that the pathogenesis of sigmoid volvulus involves not only mechanical factors but also histopathological abnormalities, in particular disturbances of the enteric nervous system.<sup>10</sup> Fujiya *et al.*<sup>10</sup> suggested that a reduction in the extent and number of enteric plexus and ganglion cells could be an important mechanism that predisposes to the torsion of the sigmoid colon.

Sigmoid volvulus represents the third leading cause of colon obstruction in adults.<sup>9</sup> This condition tends to occur in older adults (>70 years old), with a predominance of male gender.<sup>1-4,6-9,11</sup> The male predominance is associated with anatomical factors since the sigmoid colon length is significantly longer in men than in women.<sup>12</sup> Alatise *et al.*<sup>12</sup> also showed that the length and width of the sigmoid colon and the mesocolon increase with advancing age, which could explain the greater prevalence of sigmoid volvulus in elderly people.

In the majority of cases, patients with sigmoid volvulus are debilitated and institutionalized, with chronic medical conditions, and tend to have a history of chronic constipation.<sup>3</sup> However, in a recent series,<sup>11</sup> the majority (81.7%) of patients with sigmoid volvulus were independent, with only 7.9% of patients from nursing homes. The authors underlined that sigmoid volvulus is not an exclusive disease of the elderly and fragile population.<sup>11</sup> In younger patients, sigmoid volvulus could be associated with disorders of colonic motility leading to megacolon, such as Hirschsprung's disease and Chagas disease.<sup>13</sup>

Sigmoid volvulus may be acute or chronic in its presentation, with periods of exacerbation and quiescence. Almost 40% to 60% of patients have a history of prior episodes.<sup>14</sup> The probability of recurrence of sigmoid volvulus increases after the second episode and each subsequent episode represents a higher risk of bowel ischemia and perforation.<sup>1,4</sup> Mortality can be as high as 20% after recurrence of sigmoid volvulus, which emphasizes the need for a scheduled definitive prophylactic treatment.<sup>8,9</sup>

The most common presenting symptoms of non-complicated sigmoid volvulus are abdominal pain, distention and constipation (the classic triad).<sup>1</sup> The findings on physical examination are variable, depending on the degree of torsion. Hypotension and tachycardia, hypoactive or absent bowel sounds, rebound tenderness, and blood in rectal digital examination, are suggestive of complicated sigmoid volvulus with bowel ischaemia or perforation.<sup>1</sup> There are no laboratory findings diagnostic of colonic volvulus; however, a significant leukocytosis or metabolic acidosis may be suggestive of bowel compromise (ischaemia, necrosis or perforation).<sup>13</sup>

Radiographic imaging is another important step in the work-up of these patients. The sensitivity of plain abdominal radiographs for the diagnosis of sigmoid volvulus is 60%, while contrast-enhanced computed tomography (CT) has a sensitivity near to 100%.<sup>2,3</sup> Radiographic images typically reveal a distended loop twisted at an acute angle resembling a coffee bean or a bent inner tube deformity arising from the left lower quadrant toward the upper abdomen, and a decompressed colon distal to the point of occlusion.<sup>1</sup> CT is the method of choice to confirm diagnosis of sigmoid volvulus in atypical cases and allows the evaluation of the mechanism and potential complications of sigmoid volvulus.<sup>1,2,3</sup>

Endoscopy is a diagnostic and therapeutic modality in sigmoid volvulus. Endoscopic findings include a pinpoint obstruction, usually 20 to 30 cm from the anal verge, with a characteristic spiral convergence of the colonic folds, known to as the "whirl" sign. The closed-sigmoid loop is characteristically dilated. Endoscopy also allows for the assessment of colon viability. The mucosa may appear normal, or with congestion and violaceous color suggestive of ischemia, or with dark spots and blood indicative of necrosis.<sup>3</sup> Colonoscopic decompression is currently accepted as the initial step in the management of clinically stable patients with sigmoid volvulus. However, it should be regarded as a temporizing measure given the high recurrence rate of volvulus even after a successful endoscopic reduction.<sup>9</sup>

Various studies <sup>2,8,9</sup> have examined the success rate of endoscopic detorsion as the first-line treatment of uncomplicated acute sigmoid volvulus, reporting an efficacy rate that varied between 62% and 95%. No endoscopic complications were described. However, a very high recurrence rate (46%-67%)<sup>2,5,6,8,9</sup> was described in patients with sigmoid volvulus treated conservatively, comparing to those submitted to sigmoidectomy (3%),<sup>9</sup> underlining the value of prophylactic surgery after initial endoscopic decompression.

Moreover, Ifversen *et al.*<sup>5</sup> found that patients treated conservatively on their first admission had a significant poorer survival rate than those treated surgically (p=0.036), emphasizing that patients without a high operative risk should be treated with surgery after the first episode of acute sigmoid volvulus.

Thus, the currently accepted method of treatment for sigmoid volvulus is non-emergent surgery following non-operative reduction and clinical stabilization, which should be performed during the index admission or soon thereafter.<sup>1</sup> This strategy converts an emergent situation into an elective one. The major benefit of non-emergent surgery is that it allows the improvement of patient's general condition, which is particularly important in patients with serious concomitant comorbidities.<sup>15</sup>

Non-emergent prophylactic surgery after endoscopic reduction has been associated with better outcomes when compared with emergent surgery: mortality rate of 3.3% *vs.* 29%, morbidity rate of 11.4% *vs.* 34.2%.<sup>24,16</sup> Early recurrence rate was similar (0% *vs.* 0.6%).

These unfavorable outcomes support indication for emergency surgery only in cases of complicated sigmoid volvulus.<sup>1</sup> In the management of complicated forms of volvulus, laparotomy is usually performed. Restoration of intestinal continuity is not advocated in the presence of a nonviable colon or peritoneal contamination, due to the high risk of postoperative complications such as anastomotic leak and mortality.<sup>1</sup>

On the other hand, single-stage sigmoid colon resection and colorectal anastomosis is the standard of choice for the treatment of uncomplicated sigmoid volvulus.<sup>1</sup> In this context restoring intestinal continuity is associated with lower morbimortality and shorter hospital stay when compared with a two-stage operation.<sup>1,5</sup> Although stoma creation is usually not required in the nonemergency setting, the choice of surgical procedure always depends on operative findings and general patient's condition.<sup>5</sup> Comparing laparoscopy and open surgical approach, no significant differences regarding morbimortality have been identified; however, a higher recurrence rate was described for laparoscopic approach.<sup>17</sup> Moreover, the significant redundancy and mobility of the colon probably favor the open approach. So, the role and potential benefits of laparoscopic surgery in this setting are still being defined.<sup>1</sup>

After a prophylactic sigmoidectomy, the probability of recurrent sigmoid volvulus is generally low (<5%).<sup>2,4,6,8,9</sup> However, the presence of a concomitant megacolon increases the recurrence rate of volvulus from close to zero to a rate between 20-80%.<sup>18</sup> Thus, a more extensive subtotal or total colectomy is advocated as the primary technique in the presence of a megacolon, reducing the risk of post-resection recurrent volvulus.<sup>1,4,18</sup>

Although surgical treatment after non-operative reduction of a sigmoid volvulus is generally advocated, it should be noted that the favorable outcomes of this approach have been described particularly in low-risk patients (younger patients without major comorbidities). In poor surgical candidates, there is no consensus on management. In elderly patients or those with significant comorbidities, endoscopic decompression alone may be reasonable. Newer less invasive techniques, such as percutaneous endoscopic sigmoidopexy and percutaneous endoscopic colostomy, have been described in the management of patients not suitable for surgery.<sup>3</sup>

We report the case of a typical clinical picture of a sigmoid volvulus in a patient that does not present the more frequent risk factors, being a middle-age women, independent for daily living activities, without significant comorbidities except for a major predisposing factor, an idiopathic megacolon probably related to long-standing constipation.

With this case report, the authors intended to highlight the importance of pursuing a prophylactic surgical strategy in low-risk patients with recurrent sigmoid volvulus in order to prevent life-threatening complications, as well as the hazardous outcomes associated with an emergency surgery. **Corresponding author:** Raquel Alexandra Gomes dos Reis Pimentel

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# Resseção endoscópica de lipoma submucoso gigante causando obstrução cólica e prolapso anal: técnica de resseção assistida por dupla laqueação

# Endoscopic resection of a giant submucosal lipoma causing colonic obstruction and ball-valve anal prolapse: A double-ligate and resect technique

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A 53-year-old woman with Sneddon syndrome and homozygous Factor V Leiden mutation under warfarin was referred to our institution due to intermittent obstructive symptoms such as constipation, abdominal pain and distension. She also reported a mass frequently prolapsing through the anus. The physical examination, including digital rectal examination during straining, was unremarkable.

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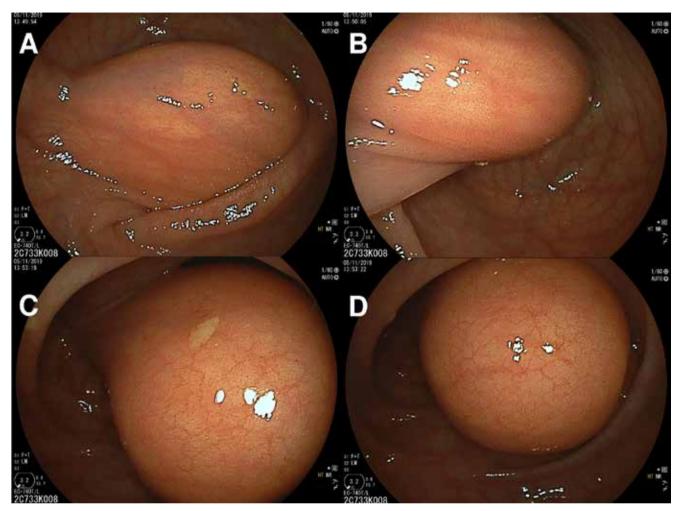
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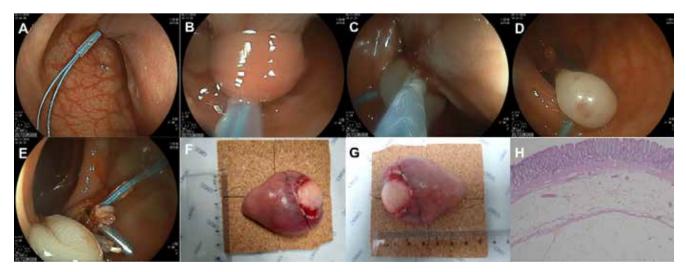
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A colonoscopy was performed, showing a 65x45mm soft yellowish mobile lesion, with a short and thick pseudo-stalk, at the distal sigmoid colon, at 25cm from the anal verge, almost obstructing the entire lumen (figure 1, A-D). After warfarin withdrawal and bridging with low-molecular-weight-heparin (LMWH), an endoscopic resection was performed using the double-ligate and resection technique: application of two 30mm endoloops® (Olympus, Tokyo, Japan) on the pseudo-stalk, followed by en bloc resection above the two endoloops® using a 27mm diathermic snare (Captivator®, Boston Scientific, Boston, USA). Immediate post-resection oozing bleeding was managed with 1:10000 diluted-adrenaline injection and three endoclips were applied on the stump. Histopathology confirmed a submucosal lipoma completely resected (figure 2, A-H). LMWH bridging was prolonged and warfarin started only after seven days. The procedure was uneventful with the resolution of patient symptoms.

Although colonic lipomas are usually asymptomatic, they can cause symptoms such as bleeding, obstruction or intussusception.<sup>1,2</sup> Prolapse through the anus has been rarely reported.<sup>3-5</sup> There is no therapeutic algorithm in the literature for the approach of symptomatic colonic lipomas. Endoscopic treatment by unroofing, dissection-based resection, endoscopic mucosal resection and loop-assisted resection showed similar clinical remission and adverse events rates, with higher resection rates for endoscopic mucosal resection and loop-assisted resection. Therefore, it has been suggested that the ideal resection technique should depend on local expertise and patient profile.<sup>6</sup>



**FIGURE 1. A-D** Endoscopic image of a 65x45mm soft yellowish mobile lesion, with a short and thick pseudo-stalk, located at the distal sigmoid colon, obstructing practically the entire lumen.



**FIGURE 2.** A-H Double-ligate and resect technique. A,B – Two endoloops<sup>®</sup> placed at the base of the pseudo-stalk. C – Endoscopic resection using a diathermic snare above the two endoloops<sup>®</sup>. D,E – Post-resection oozing bleeding at the resection site treated by combined therapy using 1:10000 diluted-adrenaline and three endoclips. F,G – Macroscopic resection specimen sizing 65x45mm. H –Adipocytes located below the muscularis mucosa (H&E 20x).

Given the hemorrhagic and thrombotic risk of our patient, a double loop-assisted resection technique was configured as a successful option, providing a definitive and safe approach. We present a successful endoscopic resection of a giant symptomatic lipoma in an anticoagulated patient complicated by colonic obstruction exhibiting ball-valve anal prolapse using a double-ligate and resect technique.

# **Statement of Ethics**

The project was subjected to the standards of good clinical practice and always complied with the ethical precepts of the Helsinki's Declaration.

### **Disclosure Statement**

The authors have no conflicts of interest to declare.

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