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Acute large bowel pseudo-obstruction due to atrophic visceral myopathy: A case report

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ABSTRACT

PURPOSE: Atrophic visceral myopathy is a pathological diagnosis characterized by atrophy of the smooth muscle layers of the viscera with intact ganglia. Rarely, it can present acutely as an intestinal pseudo-obstruction. We describe a rare case report and explore how this diagnosis can be distinguished from other forms of intestinal obstruction.

CASE DESCRIPTION: A 60-year-old male with a past medical history of hypothyroidism presented to the emergency department with a two-day history of worsening abdominal distention and pain associated with nausea and vomiting. Upon evaluation patient was found to have tachycardia, with abdominal distention and localized tenderness with peritonitis. Computed tomography demonstrated large bowel obstruction, likely caused by sigmoid volvulus. The patient underwent emergent laparotomy. Intra-operatively, the entire colon was found to be extremely dilated and redundant. With a working diagnosis of recurrent sigmoid volvulus causing intermittent large bowel obstruction, a sigmoid colectomy and primary anastomosis was performed. Pathology revealed atrophic visceral myopathy, with an extremely thin colonic wall and atrophic circumferential and longitudinal muscularis propria without inflammation or fibrosis. The ganglion cells and myenteric plexus were unaffected. Post-operatively, the patient developed prolonged ileus requiring nasogastric decompression and parenteral nutrition. The ileus resolved with pro-kinetic agents, and patient was discharged home on post-operative day fifteen.

CONCLUSIONS: Atrophic visceral neuropathy is a rare cause of intestinal pseudo-obstruction. While often presenting with chronic obstruction in younger populations, we present a rare late-onset acute presentation that may have been secondary to underlying hypothyroidism.

1. Introduction

The loss of intrinsic smooth muscle and neuronal activity within the small and large intestines impairs motility and has previously been demonstrated to be a potential cause of pseudo-obstruction of the bowel. Pseudo-obstruction has previously been defined as signs and symptoms of intestinal obstruction in the absence of a true lumen-occluding mechanical obstruction [1]. It can be differentiated from true obstruction via radiography with enteric contrast or via endoscopy.

Atrophic visceral myopathy, a relatively rare diagnosis, typically presents with chronic pseudo-obstruction symptoms. We present a case of a previously asymptomatic 60 year-old male who developed severe acute onset pseudo-obstruction. The case is presented in accordance with the SCARE guidelines for surgical case reports [2]. Prior to publication the patient provided written consent for the de-identified presentation of his clinical course.

2. Case presentation

A 60 year-old male with a past medical history of hypothyroidism (on daily levothyroxine therapy) and prior inguinal hernia repair (14 years prior) presented to our emergency department with complaints of acute onset abdominal pain, nausea, vomiting, obstipation, and abdominal distention of two days duration. On presentation, patient had tachycardia (heart rate 140 beats per minute), a temperature 37.6 °C, with regular blood pressure (136/77 mmHg) and oxygen saturation (SpO2 95% on room air). His abdominal examination was significant for distention, diffuse tenderness to palpation (worse on the left side of abdomen), and tympany. There was no noted abdominal guarding or rebound tenderness. Laboratory values drawn at the time were significant for an

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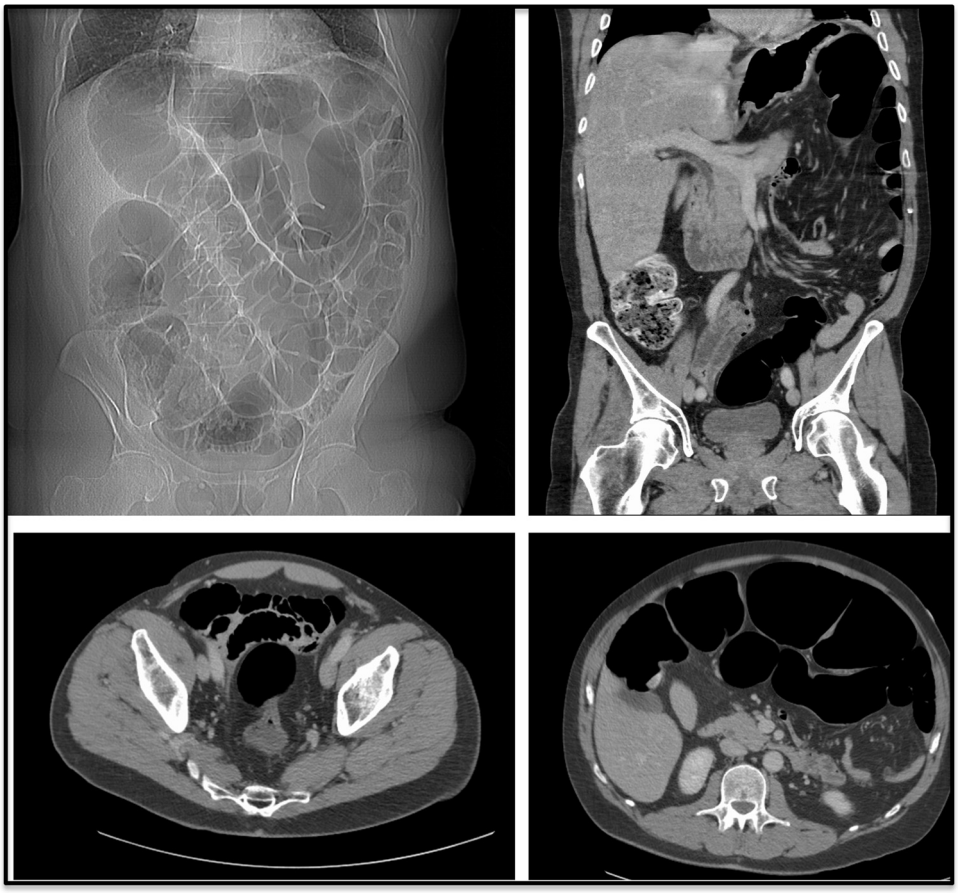

Fig. 1. Initial computed tomography (CT) abdomen and pelvis scan findings on patient presentation.

elevated white blood count of $12.53 \times 10^3/cm^2$ (89.0% neutrophils). Serum lactate was 1.7 mmol/L. A stat computed tomography (CT) scan of the abdomen and pelvis with intravenous contrast was suggestive of an acute obstruction of the sigmoid colon (Fig. 1). The imaging however did not reveal a clear transition point, was atypical for sigmoid volvulus, and there was no radiological evidence of ischemia. A rectal enema was recommended to further visualize a potential transition point, however due to the patient's worsening clinical picture, emergent operative exploration was chosen. The patient was consented and taken to the operating room for exploratory laparotomy.

A laparotomy incision was utilized in the lower midline abdomen. Upon exposure, an extremely large colon was noted (including ascending, transverse, descending, and sigmoid colon) (Fig. 2). The sigmoid mesocolon was elongated with a narrow base. No evidence of internal herniation, focal ischemic segment, or adhesive band was identified. The small bowel was run from the ligament of Treitz to the ileocecal valve, and all appeared healthy and well perfused. Based on the findings and the extremely dilated and redundant sigmoid colon, and the risk of possible intermittent sigmoid volvulus, the decision was made to perform a sigmoid colectomy with primary anastomosis. This was performed with a linear cutting stapler in a side-to-side fashion to two sized-matched regions of large bowel without tension. No diverting ostomy was created due to the satisfactory appearance of the anastomosis and negative intraoperative leak testing.

Upon evaluation by surgical pathology, the segment of resected colon (95 cm in length, 12 cm in circumference) was noted to have extreme thinning of the bowel wall throughout. Microscopically, the thin wall showed atrophic internal circumferential and external longitudinal layers of the muscularis propria, with the most severe atrophy and loss of muscle fibers identified in the external longitudinal muscle layer. Ganglion cells and the myenteric plexus appeared normal in unaffected wall, and no fibrosis was noted (Fig. 3).

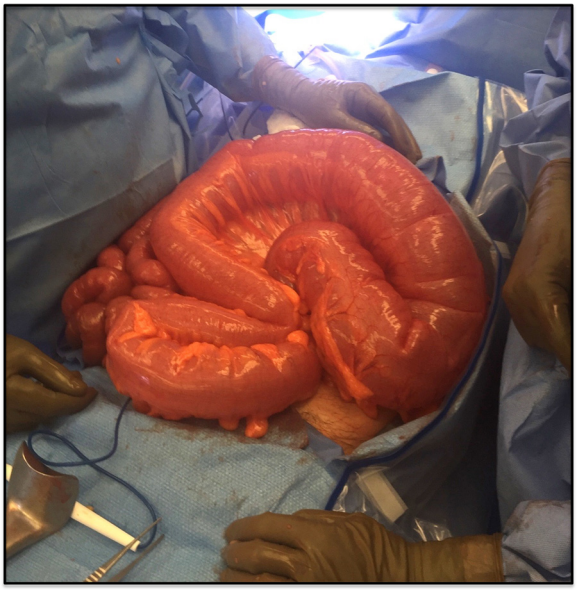

Fig. 2. Intraoperative findings and gross appearance of colon.