VOL. 12, NUM.1 (2022)

Case report

REFRACTORY ACUTE COLONIC PSEUDO-OBSTRUCTION PRECIPITATED BY MENINGIOMA AND ACUTE PONTINE MYELINOLYSIS

Pseudo-obstrução colônica aguda refratária precipitada por meningioma e mielinólise pontina aguda

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Abstract
Ogilvie´s syndrome, also defined acute colonic pseudo-obstruction, is characterized by massive dilation of the colon without mechanical obstruction. Although several neurological conditions have been described as associated with acute colonic pseudo-obstruction, there is no previous report of brain tumor and acute pontine myelinolysis precipitating such condition. This is a case report of a man diagnosed with acute colonic pseudo-obstruction precipitated by neurological disturbances. Despite the efforts, the disease was refractory to all conservative measures, requiring surgical intervention. We presented a unique case of refractory acute colonic pseudo-obstruction precipitated by meningioma and central pontine myelinolysis, leading to colonic resection and colostomy.
Keywords: Colonic Pseudo-Obstruction; Meningioma; Myelinolysis, Central Pontine

Resumo
A síndrome de *Ogilvie*, também definida como pseudo-obstrução colônica aguda, é caracterizada por dilatação maciça do cólon sem obstrução mecânica. Embora várias condições neurológicas tenham sido descritas como associadas à pseudo-obstrução colônica aguda, não há relato prévio de tumor cerebral e mielinólise pontina aguda precipitando tal condição. Este é um relato de caso de um homem diagnosticado com pseudo-obstrução colônica aguda precipitada por distúrbios neurológicos. Apesar dos esforços, a doença foi refratária a todas as medidas conservadoras, necessitando de intervenção cirúrgica. Apresentamos um caso único de pseudo-obstrução colônica aguda refratária precipitada por meningioma e mielinólise pontina central, levando à ressecção colônica e colostomia.
Palavras-chave: Pseudo-Obstrução do Colo; Meningioma; Mielinólise Central da Ponte

Introduction

Ogilvie´s syndrome is a condition characterized by clinical features of intestinal obstruction, occurring in the absence of any mechanical obstructing lesion.[1] It was first described in 1948 by Ogilvie in association with retroperitoneal malignancy infiltrating the celiac plexus.[2] *Dudley et al.* named the syndrome acute colonic pseudo-obstruction (ACPO), in recognition of the fact that the obstruction is functional rather than mechanical.[3] The pathophysiology of the disease is poorly understood, with the prevailing hypothesis being an imbalance in colonic autonomic innervation. Several underlying causes have been described that predispose to ACPO. The association with neurological diseases has been previously reported, although uncommon.[4,5] Thus, we present a case

report of a severe ACPO precipitated by meningioma and acute pontine myelinolysis. An attempt to colonic decompression was performed with temporary success. A definitive success was only obtained after surgical colostomy.

Case report

A 71-year-old man with hypertension and type 2 diabetes mellitus was transferred from another institution with a recent onset history of nausea, postprandial vomiting, diarrhea, and abdominal distention. Physical examination showed mild confusion and somnolence. The vital signs were normal (blood pressure 116/60 mmHg and heart rate 82 bpm). There was abdominal distention and tenderness and rebounding pain without bowel sounds. Laboratory tests at admission were as follows: hemoglobin 10.3 g/dL; white blood cell (WBC) count 10,500/mm3 (neutrophils 84%); platelets 301,000/mm^3; creatinine 1.08 mg/dL; sodium 165 mEq/L; potassium 2.6 mEq/L; and C-reactive protein > 12 mg/dL. Blood and urinary cultures were negative. Plain radiographs of his abdomen showed massive dilation of all colonic segments (Figure 1A). The abdominal computed tomography (CT) confirmed colonic dilation and ruled out small intestine dilation, tumors, or mesenteric ischemia (Figures 1B and 1C).

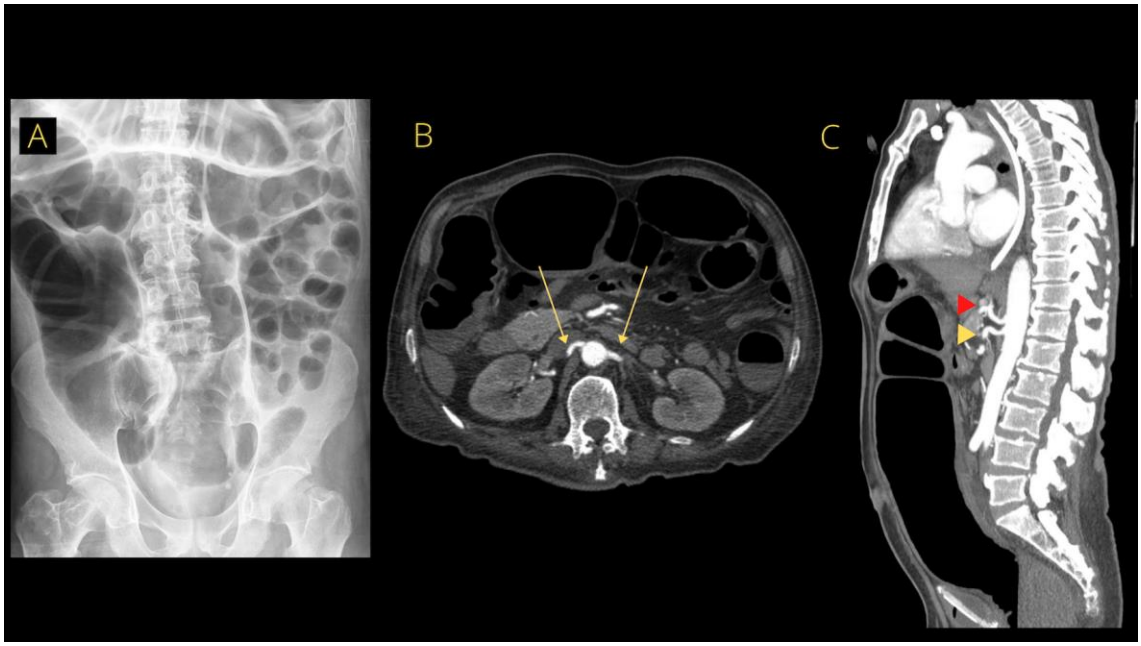

Figure 1. *Abdominal X-Ray showing marked diffuse colonic dilation (1A). The contrast abdominal computed tomography shows marked colonic dilation without evidence of mechanical obstruction (1B and 1C). There were no signs of proximal renal artery stenosis (red arrows) (1B). There was also no signs of mesenteric ischemia (1C). The superior mesenteric artery is represented by a red arrow and the celiac artery is represented by the yellow artery.*

His medical history was reviewed and was characterized by a recent admission at a general hospital due to confusion and decreased mental status. He also developed abdominal distention, diarrhea and rapid-onset hypokalemia and hypernatremia, that was not properly corrected. He received systemic antibiotics (ciprofloxacin and metronidazole) for possible abdominal infection. Since the patient got worse, he was transferred to our tertiary hospital.

At our institution, the electrolyte abnormalities were then corrected. His abdominal pain was managed without opioids. An upper gastrointestinal endoscopy revealed mild antral gastritis. A diagnosis of ACPO was performed. The abdominal distention was managed conservatively with a combination of bowel rest, nasogastric tube decompression, and rectal tube placement. Total parenteral nutrition was initiated to allow appropriate nutritional support. All electrolytes were

maintained within the normal range. After no response within 48 hours, intravenous neostigmine was started (2mg over a period of five minutes) with partial and only temporary improvement in colonic distention. Two additional attempts with neostigmine and use of oral polyethylene glycol were made without success in the following days.

Due to persistent somnolence, a head CT was performed showing a large bi-frontal mass (4.0 x 3.9 x 4.6 cm) with surrounding edema compatible with meningioma. There were also signs of ischemia in the posterior inferior cerebellar artery and findings compatible with central pontine myelinolysis **(Figure 2A/B)**. A neurological consultation was performed. Since the patient and family deferred surgical removal of the tumor, systemic corticosteroids were administered for reducing brain edema. After seven days, his clinical status and somnolence improved, the brain CT confirmed edema reduction **(Figure 2C)**, but the abdominal distention persisted.

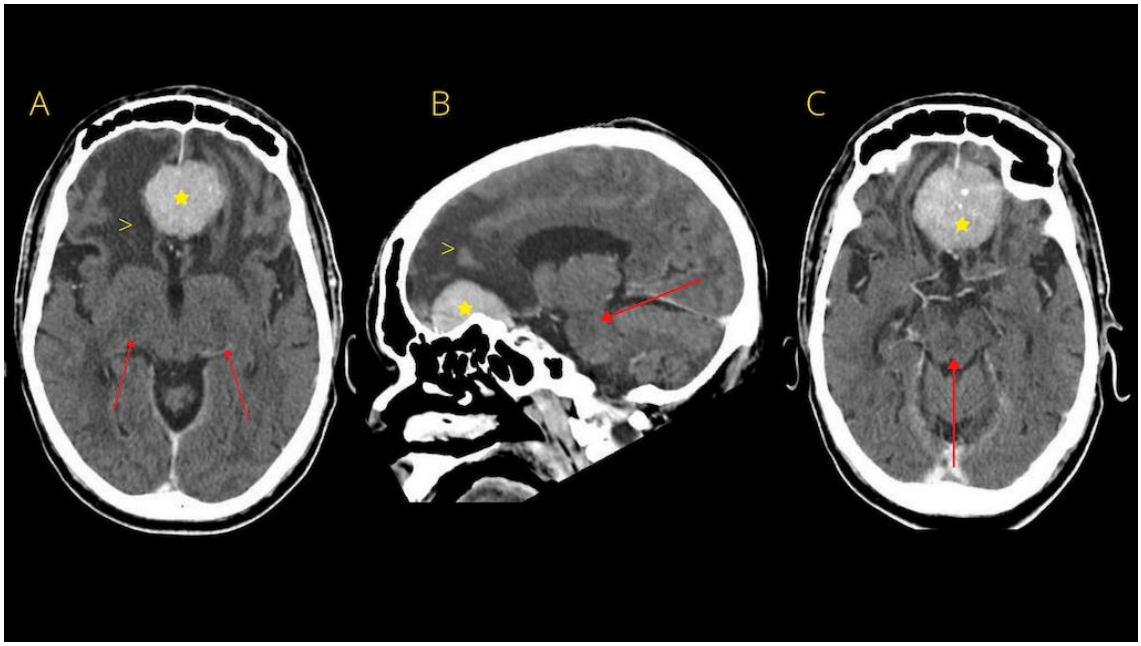

Figure 2. *Noncontract brain CT-scan showing a large enhancing bifrontal mass (yellow star) with surrounding vasogenic edema (yellow arrowhead) (2A). The lesion affects the base of the skull (2B). Contrast enhanced CT after seven days of systemic corticosteroids (2C): there is persistence of large enhancing bifrontal mass with internal calcification and marked reduction of brain edema. All images show centrally located region of low attenuation within the pons (red arrows) compatible with central pontine myelinolysis.*

A colonoscopic decompression was attempted with initial success **(Figure 3A)**. However, the abdominal distention recurred after some days. Finally, a decompressive surgical colostomy was performed with success **(Figure 3B)**. Surgical biopsy revealed colonic mucosa with mild distortion of glandular architecture **(Figure 3C)** and normal myenteric plexus **(Figure 3D)**.

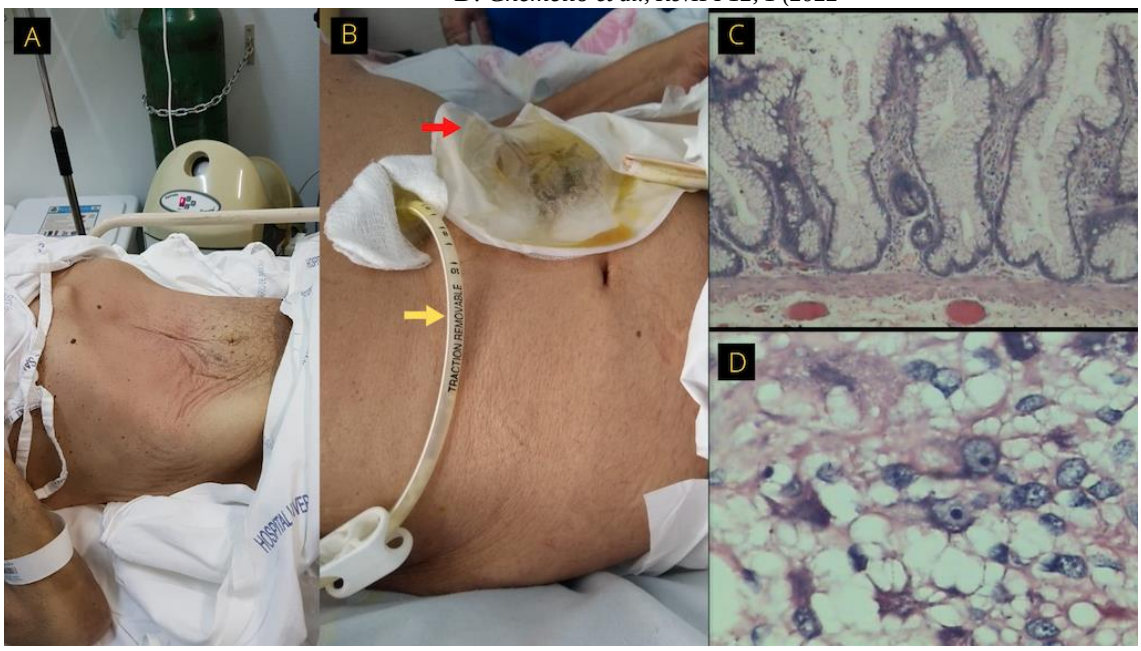

Figure 3. *Immediate results after colonoscopic decompression, showing temporary improvement of the colonic distention (3A). Result after surgical intervention (3B). The colostomy bag is shown (3B, red arrow), as well as the enteral tube (3B, yellow arrow). Histological analysis of the colon revealed colonic mucosa with only mild distortion of glandular architecture (3C) and normal myenteric plexus (3D) (Hematoxylin and eosin stain).*

After some days, the patient was able to restart enteral nutrition. He was discharged to the home-care unit. The 12-month follow-up showed normal status, without further abdominal distention.

Discussion

We present a unique case report of a patient who developed ACPO precipitated by several neurological disturbances. Despite the efforts, the disease was refractory to all conservative measures, necessitating surgical colonic decompression.

Ogilvie´s syndrome is uncommon, with a reported incidence of approximately 100 cases per 100,000 inpatient admissions.[6] Altered extrinsic regulation of colonic function by the sympathetic and parasympathetic nervous system is the most commonly mechanism suggested for the disease.[7]

In the present case, we hypothesized the presence of a large meningioma and brain edema in an elderly male lead to altered mental status and electrolyte disturbances. The rapid-onset hypernatremia precipitated central pontine myelinolysis. All these abnormalities caused bowel neurological imbalance and refractory ACPO. Previous reports have described neurological disease as a predisposing factor in 9.3% of cases of Ogilvie´s, including meningoencephalitis.[5,8] However, as far as we are concern, this is the first case report of Ogilvie´s syndrome potentially precipitated by meningioma and central pontine myelinolysis.

Regarding treatment, supportive therapy is the preferred initial management and should be instituted in all patients. These measures include given nothing by mouth, intravenous fluid administration, and correction of electrolyte imbalances.[4] For refractory cases the best pharmacological treatment is intravenous neostigmine, which leads to prompt colon decompression in the majority of patients after a single dose.[4,9] For those who have failed supportive and pharmacologic therapy with neostigmine, colonic decompression is the initial invasive procedure of

choice. It´s usually indicated for those with marked cecal distention (>10 cm) of significant duration (3-4 days). Colonoscopy is performed to prevent bowel ischemia and perforation and should not be performed if overt peritonitis or perforation is present.[4] Finally, after failure with all mentioned attempts, a cecostomy or colostomy is the final option.

The surgical intervention is rarely necessary and should be reserved for patients with ischemia, peritonitis, or ACPO refractory to decompression via pharmacologic or medical therapy.[10] Percutaneous tube cecostomy was once considered the standard of care with success rates up to 95%. However, due to high complication rates (related to the management of the stoma, infection, and dislodgement of the tube) led to loss in popularity. As such, most services prefer colostomy with or without colonic resection.[11] In the present case, the option for colostomy was mainly based on limited surgical experience with cecostomy, attempting to avoid postoperative complications.

One could argue that a neurosurgical intervention of the meningioma would be effective in improving the abdominal status. However, this option was considered extremely aggressive. Also, the patient and his family deferred any neurosurgical intervention. Finally, it would be impossible to predict the consequences of another neurological intervention in the colonic motility.

The main prognosis of ACOP is not established. In a retrospective single center cohort of patients with Ogilvie´s syndrome, *Haj et al.* showed the disease is associated with low inpatient mortality. More interestingly, there was no clinical benefit that interventional measures (neostigmine, colonoscopy or surgery) lead to improved outcomes compared to a more conservative strategy.[12] These facts reinforce the rarity of our case.

References

1. Georgescu EF, Vasile I, Georgescu AC. Intestinal pseudo-obstruction--a rare condition with heterogeneous etiology and unpredictable outcome. A case report. *J Gastrointestin Liver Dis.* 2008;17(1):77-80.
2. Ogilvie WH. William Heneage Ogilvie 1887-1971. Large-intestine colic due to sympathetic deprivation. A new clinical syndrome. *Dis Colon Rectum.* 1987;30(12):984-987.
3. Dudley HA, Sinclair IS, Mclaren IF, Mcnair TJ, Newsam JE. Intestinal pseudo-obstruction. *J R Coll Surg Edinb.* 1958;3(3):206-217.
4. Saunders MD, Kimmey MB. Systematic review: acute colonic pseudo-obstruction. *Aliment Pharmacol Ther.* 2005;22(10):917-925.
5. Vanek VW, Al-Salti M. Acute pseudo-obstruction of the colon (Ogilvie's syndrome). An analysis of 400 cases. *Dis Colon Rectum.* 1986;29(3):203-210.
6. Ross SW, Oommen B, Wormer BA, et al. Acute Colonic Pseudo-obstruction: Defining the Epidemiology, Treatment, and Adverse Outcomes of Ogilvie's Syndrome. *Am Surg.* 2016;82(2):102-111.
7. Wells CI, O'Grady G, Bissett IP. Acute colonic pseudo-obstruction: A systematic review of aetiology and mechanisms. *World J Gastroenterol.* 2017;23(30):5634-5644.
8. Lin W-C, Chu C-H, Shih S-C, Cheng S-J. Acute colonic pseudo-obstruction as a presenting feature of meningoencephalitis. *Journal of Acute Medicine.* 2016;6(3):70-72.
9. Ponec RJ, Saunders MD, Kimmey MB. Neostigmine for the treatment of acute colonic pseudo-obstruction. *N Engl J Med.* 1999;341(3):137-141.

10. Jain A, Vargas HD. Advances and challenges in the management of acute colonic pseudo-obstruction (ogilvie syndrome). *Clin Colon Rectal Surg.* 2012;25(1):37-45.

11. Galban D, Baiel JJ. An Alternative Approach to the Terminal Management of Ogilvie Syndrome. *Case Rep Gastroenterol.* 2017;11(2):352-358.

12. Haj M, Rockey DC. Ogilvie's syndrome: management and outcomes. *Medicine (Baltimore).* 2018;97(27):e11187.