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LETTER TO THE EDITOR

# Relapsing paralytic ileus in multiple sclerosis requiring surgery: a video case report

G. Chiaro[1,2] · C. Fratila[1] · F. Martig[3] · C. Zecca[1] · C. Gobbi[1]

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Dear Editors,

Autonomic dysfunction has been previously described in patients with multiple sclerosis (MS), with prominent involvement of the cardiovascular, urinary, and gastrointestinal branches of the autonomic nervous system (ANS) [1–3]. Gastrointestinal dysfunction is particularly remarkable in these patients, with both constipation and fecal incontinence being its most prevalent manifestations [4, 5]. Moreover, correlations between disability, disease duration, prevalence, and severity of bowel symptoms have been drawn [6]. Recently, acute manifestations of gastrointestinal dysfunction, ranging from acute dysphagia to gastroparesis to acute paralytic ileus have been increasingly recognized and incorporated into patients' assessments [4].

We had the chance to witness the case of a 44-year-old woman with a clinically stable relapsing–remitting form of MS, a mild functional disability (EDSS 2.0 defined by relevant fatigue), treated with interferon beta-1a 44 mcg (Rebif®) for the last 5 years. She was admitted to the emergency department because of sudden abdominal pain, nausea, and vomiting. She had last passed stools 2 days before; her medical history was otherwise negative. Clinical examination was remarkable for epigastric tenderness, with no signs of peritoneal irritation. A detailed review of the ANS utilizing the Italian version of the Composite Autonomic Symptom Scale-31 (COMPASS-31) [7] disclosed selective involvement of the enteric nervous system in terms of constipation. Orthostatic intolerance as well as pupillomotor, vasomotor, secretomotor, or bladder dysfunction were absent. A simple supine versus standing blood pressure (BP) measurement excluded orthostatic hypotension or postural tachycardia. A thorough neurological examination did not show any other sign of AF.

Routine laboratory tests (inflammatory indices, liver, biliary tract and kidney functions, lactate, and creatine phosphokinase) were all normal. An abdominal CT scan showed signs of small bowel obstruction with a possible beak and feces sign. As there were no clear signs of bowel wall suffering or free fluid collection, a conservative treatment was attempted, but the patient soon showed fecal vomiting. An emergency diagnostic laparoscopy did not find any mechanical obstruction but it did show an atonic bowel with a complete absence of peristalsis (see Video sample 1 in the Electronic supplementary material). Intraoperative biopsies yielded negative results. The clinical course was free from complications and the patient was discharged home 4 days later. After 5 days, she consulted our emergency department for recurrence of the same signs and symptoms. Lab tests, an abdominal X-ray, and a CT scan confirmed the same diagnosis of paralytic ileus without mechanical obstruction, which resolved after treatment with amidotrizoic acid (Gastrografin®) and neostigmine. A brain 3T-MRI showed a nonenhancing lesion anteriorly in the lower medulla, and a spinal 3T-MRI disclosed multiple cervical and dorsal demyelinating lesions (at the C1-2-3-4 and T8-9 levels) without gadolinium enhancement (Fig. 1), with a stable lesion load

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✉ C. Gobbi
claudio.gobbi@eoc.ch

[1] Neurocenter of Southern Switzerland, Civic Hospital of Lugano, Via Tesserete, 48, 6900 Lugano, Switzerland

[2] Department of Biomedical and Neuromotor Sciences, University of Bologna, Bologna, Italy

[3] Department of Surgery, Civic Hospital of Lugano, Lugano, Switzerland

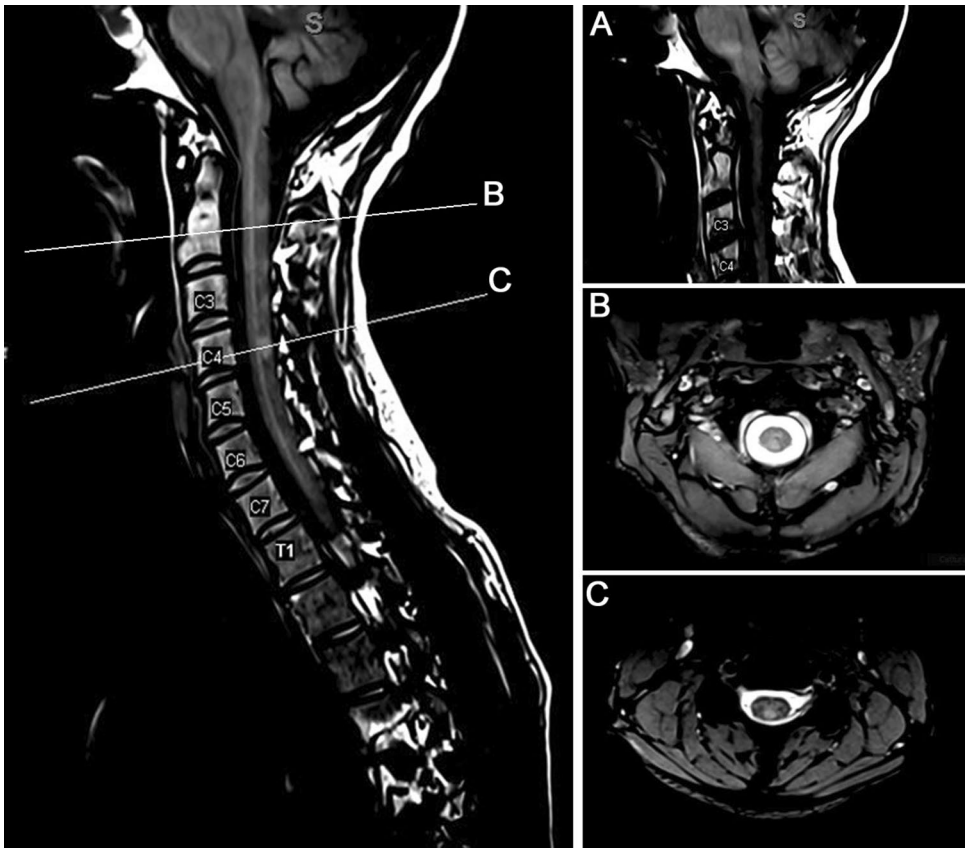


**Fig. 1** T2-weighted 3-tesla MRI sequences showing non-enhancing lesions in the anterior bulbo-medullary junction (A), (B) anterior portion of the medulla at C2 and (C) C4 levels. Smaller lesions are scattered throughout the cervical spine

compared to the year before. Imaging of the lower spine was also performed, which did not show any pathological finding. The patient was discharged 4 days later.

This case can be added to a growing number of reported cases of acute paralytic ileus in MS patients. Given that infectious, compressive, and structural causes for the ileus were excluded, we argued that a relationship between a relapse of MS and the paralytic ileus could be the most plausible cause. Historically, it has always been postulated that AF in MS patients is likely due to plaques distributed throughout the brainstem and spinal cord affecting anatomically widespread autonomic regulatory areas and their connections [3, 8, 9], and that the level of AF correlates to MS disease severity [10]. It is known that brainstem lesions, especially those affecting the locus coeruleus, account for most of the sympathetic dysfunction in MS [11]. Although no new active lesions were detectable at the spinal MRI, we believe there is persuasive evidence that MS is the underlying cause. It is common for pre-existing MS lesions to become more symptomatic under a variety of conditions. Recent studies, though, are shifting the attention towards a possible concomitant peripheral involvement in MS, with the suggestion being that demyelination occurs not only within the central but also along the peripheral nervous system, as shown in early neuropathological reports [12]. To this end, an absent or impaired response to the quantitative sudomotor axon reflex test (QSART) has recently been used as a marker of postganglionic (and preganglionic) axonal damage in MS [11, 13]. A proof-of-concept study has visualized and quantified peripheral nerve lesions in MS in vivo through high-resolution MRI studies [14]. Albeit fascinating, and surely worth deeper investigation, as some parameters of peripheral AF might even help to distinguish between MS phenotypes, the aforesaid studies did not provide evidence of enteric nervous system involvement, which is not equivalent to peripheral nervous system involvement.

It could be argued that more peripheral lesions that were invisible to routine neuroimaging might have played a role in precipitating the two described episodes of paralytic ileus by misbalancing a preexisting precarious equilibrium within the documented spinal lesion load. Peripheral involvement in our case remains highly speculative. Neurologists should be aware that MS might cause acute and severe impairment of the autonomic system, particularly in those patients with a previous history of dysautonomia and inflammatory spinal lesions.

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