

Takao Nagashima · Masahiro Iwamoto · Seiji Minota

Semiquantitative assessment of the intestinal motility in chronic intestinal pseudo-obstruction in systemic sclerosis and mixed connective tissue disease by Sitzmarks capsule

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Chronic intestinal pseudo-obstruction (CIPO) is a rare complication of connective tissue diseases, especially systemic sclerosis.^{1,2} Treatment of CIPO remains problematic. Surgical resection of enlarged bowel was shown to be unsatisfactory. Medical treatment of CIPO with metoclopramide, domperidone, cisapride, erythromycin, and octreotide (a long-acting somatostatin analogue) alleviates the symptoms only in its initial stage. The clinical diagnosis of CIPO is not difficult considering past medical histories in which repeated episodes of abdominal obstructive symptoms were resistant to treatment, and radiographic findings of enlarged intestine. The correct diagnosis needs gastrointestinal manometry, but this is only available in limited referral centers. Radioisotope gastric emptying studies are easily feasible, although assessment of the small intestine is quite difficult. Barium examinations are not recommended for those patients suspected of severe colonic involvement because they may cause life-threatening impactions.³ The Sitzmarks capsule (Konsyl Pharmaceuticals, Fort Worth, TX, USA) is a diagnostic tool for the evaluation of intestinal motility in constipated patients. It contains 20 radiopaque markers, 4.5 mm in diameter and 1.0 mm in thickness, and is made of a mixture of barium sulfate and polyvinyl chloride. The Sitzmarks diagnostic test helps physicians select the best therapeutic options for severe constipation. Their simplified protocol is to administer the capsule at day 0 and obtain a flat abdominal X-ray at day 5 to see how

many markers remain in the gastrointestinal tract and where they are located. Patients who expel at least 80% markers have grossly normal colonic transit. Colonic transit delay has been demonstrated in a patient with systemic sclerosis by using similar material.⁴ It has also been adopted for detecting diabetic and other gastrointestinal motility disturbances.^{5,6} Here, we report two cases of CIPO in collagen vascular diseases, whose intestinal motility was evaluated using Sitzmarks.

Case 1

A 62-year-old woman, diagnosed with mixed connective tissue disease 8 years ago, was admitted to our rheumatology unit, complaining of abdominal pain and frequent diarrhea. The diagnosis of mixed connective tissue disease was made from Raynaud's phenomenon, sclerodactyly, arthritis, lymphadenopathy, and positive anti-U1-RNP antibody. She had been admitted repeatedly because of her digital and lower leg ulcer. Three years previously, prominent esophageal enlargement and steatorrhea had been pointed out. A clinical diagnosis of CIPO was made from her abdominal distension and frequent diarrhea 1 year previously. She could take low-residue meals with cisapride and antibiotics at that time. An additional diagnosis of ischemic colitis was made from multiple ulcers and hemorrhage in her left hemicolon by colonoscopy. In spite of the administration of intravenous antibiotics and fasting therapy for more than 1 month, her abdominal pain continued. Abdominal plain roentgenography (X-ray) and computed tomography (CT) demonstrated a marked enlargement of small and large intestine. To evaluate the intestinal motility, she was given a Sitzmarks capsule; all 20 radiopaque rings remained in her stomach 5 days after ingestion. Even after 1 month, only two rings had moved out of her stomach into the intestine. The prokinetic drug trial was abandoned because of recurrent bouts of abdominal pain. She was finally discharged after hyperalimentation through a central vein was implemented.

T. Nagashima (✉) · M. Iwamoto · S. Minota
Division of Rheumatology and Clinical Immunology, Department of
Medicine, Jichi Medical University, 3311-1 Yakushiji, Shimotsuke
329-0498, Japan
Tel. +81-285-58-7358; Fax +81-285-44-2779
e-mail: naga4ma@jichi.ac.jp

Case 2

A 66-year-old woman, diagnosed with systemic sclerosis (SSc) at the age of 45, was admitted to the gastrointestinal unit of our hospital due to nausea and abdominal pain. She had been treated with D-penicillamine and prednisolone by her family physician, but the details were unknown. She had interstitial pneumonia, mild pulmonary hypertension, and reflux esophagitis. Her abdominal symptoms were considered initially to be due to a mechanical obstruction, because she had had a hysterectomy. After her abdominal pain subsided, a semi-liquid diet was initiated; her abdominal symptoms recurred a few days later. Neither metoclopramide nor dinoprost improved her bowel movement. Because no mechanical obstruction or stenosis was found by gastroendoscopy, colonoscopy, and total small intestinal fiberoptic, to account for the marked enlargement of intestine by abdominal X-ray and CT, she was considered to have CIPO as a complication of SSc. She was transferred to our rheumatology unit for treatment of CIPO. She was given one Sitzmarks capsule, and an abdominal X-ray was taken 5 days later. Although all 20 rings were dispersed throughout her abdomen, no ring was excreted (Fig. 1). Erythromycin (EM) 100mg three times a day caused four rings to be excreted after 5 days. Semi-liquid to low-residue diet did not induce abdominal pain or bloating sensation as long as EM was administered concomitantly, and she had a regular fecal frequency. After all rings had been confirmed as excreted, a Sitzmarks capsule was readministered. Only four rings remained in her abdomen on the X-ray taken 5 days after ingestion.

Discussion

While the diagnosis of CIPO had already been made 1 year before the Sitzmarks test in case 1, the test was done soon after the diagnosis was made in case 2. Needless to say, pharmacotherapy for CIPO is only effective before smooth muscle atrophy and fibrosis develop. The Sitzmarks test was shown here to be safe, easy, and convenient for the evaluation of intestinal bowel movement. The Sitzmarks test has been used mainly for assessment of severe constipation and no serious complications have been found under such conditions. We think it a highly safe procedure considering the fact that nothing happened in case 1 – all Sitzmarks capsules remained in the patient's intestine even after 1 month. Thus, the Sitzmarks test is useful in evaluating CIPO semiquantitatively and in assessing the appropriate drugs for its treatment, rather than the diagnosis of CIPO per se, which can usually be made qualitatively.



Fig. 1. An abdominal X-ray taken 5 days after Sitzmarks ingestion. Most of the radiopaque rings were observed in the patient's right abdomen (arrowheads)

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