ONLINE EXCLUSIVE

THE PATIENT

85-year-old woman

SIGNS & SYMPTOMS

– Insomnia
– Abdominal discomfort
– Urge to move at night

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THE CASE

An 85-year-old woman with hypertension presented to our hospital with a 10-month history of insomnia along with abdominal discomfort. Several months prior, the patient had undergone an esophagogastroduodenoscopy, the results of which were normal, and had received diagnoses of psychogenic insomnia and abdominal pain from her previous physician. At that time, she was prescribed eszopiclone, but her insomnia did not improve. She did not complain of any other gastrointestinal symptoms.

On examination at our hospital, the patient's abdomen was soft and nontender. Laboratory results were unremarkable. Abdominal computed tomography was performed to exclude obvious malignancy and showed no remarkable findings.

Additional history taking and physical examination were performed. The patient reported that she could sleep for only about 2 hours per night due to persistent severe discomfort around the umbilicus, which she described as "itching." The discomfort occurred along with an urge to move while she laid in a state of relaxed wakefulness. This discomfort occurred no matter what position she laid in and improved if she walked or tapped around the umbilicus for a while. She denied any unusual or uncomfortable sensations in her lower extremities.

Her symptoms were absent during the daytime and not related to diet. Furthermore, she did not have any symptoms of anxiety and/or depression; a detailed neurologic examination, including cognitive assessment and extrapyramidal system, yielded unremarkable findings. Additional laboratory tests showed a mild iron deficiency (ferritin, 52.6 µg/L; iron, 10.7 µmol/L) without anemia.

THE DIAGNOSIS

Given the patient's presentation and clinical history, the differential diagnosis included restless abdomen (which is a spectrum or a phenotypic variant of restless legs syndrome [RLS]) and its mimics, which include fibromyalgia and gastrointestinal tract diseases. We considered the characteristic symptoms of this case (ie, irresistible symptoms, lengthy duration of symptoms, and sleep problems) to better support the diagnosis of restless abdomen than its mimics.[1] In particular, abdominal discomfort that led to insomnia was characteristic of restless abdomen, helping to pinpoint the diagnosis.

DISCUSSION

RLS is a common sensorimotor disorder that is characterized by an unpleasant urge to move the legs.[2] RLS may manifest as an idiopathic condition, or it can be secondary to medical conditions such as iron deficiency and Parkinson disease.[3,4] Because the unpleasant symptom is exacerbated in the evenings, patients with RLS frequently complain of sleep disturbance.

Cases of RLS-like sensory disorders, with symptoms involving sites other than the lower

extremities (eg, arms, mouth, trunk, and genitals) recently have been reported.[5-7] Among them is restless abdomen, a rare disorder that manifests with a restless abdominal sensation and worsens the quality of sleep and life.[6]

Restless abdomen meets all other diagnostic criteria for RLS except for the affected anatomy.[6,8] In most cases of restless abdomen, the uncomfortable sensation involves the abdomen, as well as other parts of the body (eg, legs and arms). Cases in which the symptoms are confined to the abdomen are rare, with only 7 reported to date.[6,8-10] All of these cases have involved patients older than 40 years.[6,8-10]

Treatment is straightforward, but consider iron supplementation, as well

Because RLS or its variants degrade the quality of life and sleep in patients,[3,4] appropriate therapy must be initiated early. Although the optimal treatment strategy for restless abdomen is yet to be established, an oral dopamine agonist—specifically, pramipexole—has been used successfully in almost all cases.[6,8-10]

Although the optimal treatment strategy for restless abdomen is yet to be established, an oral dopamine agonist—pramipexole—has been used successfully in almost all cases.

Previous clinical research has shown that patients with RLS have low levels of iron in the brain and may benefit from iron supplementation, even if they are not anemic.[3,4] Iron replacement is suggested for patients with RLS whose fasting serum ferritin level is ≤ 75 µg/L.[4] It is not known to what extent iron deficiency is involved in the pathophysiology of restless abdomen, and further research is required to determine the optimal therapy for it.

Our patient was started on oral supplementation with sodium ferrous citrate (50 mg/d) based on an initial suspicion that iron deficiency was the cause of her restless abdomen. We also suggested that the patient undergo a fecal occult blood test or colonoscopy, but she declined because of her advanced age.

After 2 months of iron supplementation, the patient's serum ferritin levels improved (100 µg/L) and her insomnia and abdominal discomfort improved a bit. However, 3 months after starting on the iron supplementation, her symptoms flared again.

We then prescribed pramipexole 0.25 mg/d. The patient's symptoms subsequently resolved, and she no longer experienced insomnia. This favorable response to dopamine agonist therapy supported the diagnosis of restless abdomen. The patient continues to take the pramipexole to prevent a relapse.

THE TAKEAWAY

Insomnia is a common presenting complaint in primary care and sleeping pills may be prescribed without adequate investigation of the cause. However, some patients may have serious underlying diseases.[11]

Although restless abdomen is a disorder that causes severe sleep disturbance and impairs the patient's quality of sleep and life, it is not widely recognized by clinicians and may be misdiagnosed. When recognized, insomnia due to restless abdomen can be relieved by a simple therapy: oral dopamine agonists. Therefore, primary care physicians should consider restless abdomen as a potential cause of insomnia with abdominal symptoms. JFP

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