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Letter to the Editor

Abdominal paresthesia resembling restless legs syndrome successfully treated with iron supplement therapy: A case report

Restless legs syndrome (RLS) is a sensorimotor disorder characterized by the urge to move or an unpleasant sensation in the legs. Recently, some cases in which paresthesias with the urge to move body parts other than the legs have been reported [1–3]. These cases have been assumed to be phenotypic variants of RLS and successfully improved with dopaminergic medications, the most common treatment choice for RLS. Here, we report the first case with abdominal paresthesia resembling RLS, which was successfully treated with iron supplement therapy.

1. Case report

In April 2011, a 54-year-old woman underwent a cholecystectomy together with gallbladder bed resection for cholecystitis. Although the postoperative course was generally satisfactory, she started to complain of intolerable discomfort around the abdomen after the operation. Abdominal imaging was unremarkable. No remarkable laboratory data were found except for some mild abnormalities, such as a low hemoglobin level (95 g/L), elevated liver enzymes (AST 88 U/L, ALT 97 U/L) and a high concentration of C-reactive protein (44 mg/L). The discomfort in the abdomen was not ameliorated by painkillers and persisted for one month.

In November 2011, she was found to have pancreatic cancer with superior membrane artery invasion and started to undergo chemotherapy with gemcitabine. One week after treatment initiation, the intense unpleasant feeling in the abdomen returned, and she gradually developed severe sleep onset insomnia and peculiar behaviors after retiring, including jumping on the bed, making movements similar to sit-ups, and pressing an ice-pack onto her stomach. A comorbid psychiatric disorder, such as psychosis or delirium, was suspected and she was referred to the department of psychiatry. However, no signs or symptoms of psychiatric disorders were found. She reported that the discomfort was intolerable and it "drove her out of her mind". The uncomfortable sensation usually began during periods of rest and worsened in the evening. It made her feel an irrepressible urge to move and was temporarily relieved by the foregoing movements. Although she did not complain of any abnormal sensations in the legs, these behavioral findings appeared to be comparable to those associated with RLS. Her serum ferritin was normal (240 pmol/L), but her serum iron was low (7.7 μmol/L). We suspected that iron deficiency caused the unpleasant feeling and started administering sodium ferrous citrate at 100 mg/day. Two weeks later, her serum iron level was completely normalized (20.0 μmol/L), and the unpleasant abdominal sensation had been abolished.

2. Discussion

Had our patient's legs, rather than her abdomen, been affected, she showed all the essential features of RLS [4]: 1) an urge to move her body caused by an unpleasant sensation, 2) a condition that worsened during periods of rest, 3) the feeling was partially relieved by movements, and 4) the condition worsened in the evening. The leg symptoms seem to be a diagnostic prerequisite when the disorder involves body parts other than the legs. Recently, however, the concept of "RLS equivalents", in which paresthesias resembling RLS have occurred in the arms, abdomen or face without any involvement of the legs, has been proposed based on the accumulation of some case reports [1–3]. In this regard, our patient may also be classified as having a syndrome comparable to an abdominal RLS equivalent.

In our case, iron deficiency was suspected to be the cause of the symptoms, but her serum ferritin level was normal. Some cancers can elevate serum ferritin concentrations [5], which may provide an explanation for her normal serum ferritin level while she had an actual iron deficiency. Dopaminergic agents are now considered the first-line treatment for RLS [4], and RLS equivalent cases previously reported have also shown good responses to dopaminergic agents [1–3]. In RLS cases with iron-deficiency, iron supplement therapy is also recommended because brain iron deficiency is associated with RLS symptoms [4]. The abdominal paresthesia of our case was eliminated with iron supplement therapy, which further supports the possibility that paresthesias with urges to move body parts other than the legs are phenotypic variants of RLS.

Conflict of interest

The authors declare that there are no conflicts of interest.

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