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LETTER TO THE EDITOR

Reversible stiff person syndrome presenting as an initial symptom in a patient with colon adenocarcinoma

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To the Editor,

An 82-year-old previously healthy woman complained of neck and upper back heaviness and stiffness for two weeks. The heaviness and stiffness spread to the lower back and lower limbs without upper limb involvement, and caused difficulties in walking and standing. She also had trouble in opening her mouth and swallowing food. She was sent to our emergency room due to painful spasms in her back and legs. A neurological examination revealed increased muscle tone with stimulation-sensitive spasms of her lower limbs. Serum tests were normal except for an elevated creatine phosphokinase level (248 IU/L, normal range: 38–234 IU/L). Needle electromyography revealed continuous motor unit activity, which was suppressed after administration of intravenous diazepam. We made various diagnoses under the impression of a movement disorder emergency presenting with stiffness [1]. The patient did not have a drug or toxic exposure history. Neither trauma nor vaccine injection history could be tracked. As a result, neuroleptic malignant syndrome, serotonin syndrome, strychnine intoxication, and tetanus were less likely to be the diagnosis. Further evaluations of cerebrospinal fluid analysis and brain computed tomography showed no evidence of central nervous system infection or subarachnoid hemorrhage. Suspected hypocalcemia was also excluded due to a normal serum calcium level (8.5 mg/dl, range: 8.5–10.5 mg/dl). From the results of the above detailed examinations, the patient's symptoms and reasonable conjecture, our diagnosis was stiff person syndrome.

In previous reports in the literature, autoimmune diseases, endocrine diseases, and cancers have been reported as possible etiologies of stiff person syndrome [2–4]. Endocrine studies (including prolactin, cortisol, adrenocorticotropic hormone, thyroid-stimulating hormone, and free T4) in our patient were normal. We checked the serum tumor markers, and elevation of carcinoembryonic antigen (CEA) level (5.59 ng/ml, normal range $<$ 5 ng/dl) was noted. Tracking back through her history, no body weight loss, bowel habits change, abdominal discomfort, or hematochezia were noted. A stool occult blood test was negative. Colonofiberoscopy revealed a sigmoid colon polypoid lesion and adenocarcinoma was diagnosed via tissue biopsy. Left hemicolectomy was performed, and sigmoid colon adenocarcinoma, stage 2A (T3N0M0) was confirmed. Due to the stage 2A sigmoid colon cancer, she did not receive any adjuvant chemotherapy [5]. The follow-up serum CEA levels one month and six months after the operation returned to normal limits. There was no evidence of tumor recurrence after the tumor resection. The patient`s stiff person syndrome responded well to daily diazepam 6 mg therapy, and she recovered completely three months after tumor resection. It was reasonable to speculate about a relationship between colon cancer and stiff person syndrome in this patient. Thus, we believed it to be a paraneoplastic syndrome.

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Stiff person syndrome is a rare neurologic disorder first identified by Moersch and Woltmanand in 1956 [6]. The main manifestations of stiff person syndrome are fluctuating muscular stiffness and spasms. Symptoms usually progress slowly within several weeks or months. With the progression of the disease, an increase of muscle tone occurs which becomes constant.

Several diseases have been reported to be paraneoplastic syndromes of colon cancer, such as ANCA-associated vasculitis [7] and acrokeratosis paraneoplastica [8]. Paraneoplastic stiff person syndrome is usually associated with breast cancer, small cell lung cancer, and Hodgkin lymphoma [9]. However, to the best of our knowledge, paraneoplastic stiff person syndrome presenting as an initial clinical manifestation of colon cancer has never been documented. Physicians should be aware of the syndrome and perform screening examinations to detect any underlying early-stage curable malignant disease.

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