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Case Report

Unilateral chorea secondary to hyperthyroidism: a case report

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Abstract: Chorea, an abnormal involuntary movement disorder, could be associated with hyperthyroidism. Most hyperthyroid chorea cases have been reported in young ladies and rarely in elder male patients. This study reports on a 62-year-old male patient with unilateral chorea secondary to hyperthyroidism, which was proven by sonography and thyroid function blood tests. He was also diagnosed with lacunar cerebral infarction, atrial fibrillation and type II diabetes, based on brain imaging, UCG and other laboratory tests. After 2 weeks of anti-thyroid therapy, his chorea notably improved. The patient's diagnosis and treatment procedure are described here, including a literature review.

Keywords: Chorea, hyperthyroidism, involuntary movement

Introduction

Thyroid metabolism is fundamentally involved in development and functioning of both central and peripheral nervous systems. Genetic or acquired thyroid disorders may incur neurological dysfunctions, such as dystonia, dyskinesia, cognitive impairment, depression, anxiety, muscle weakness, tremor, or chorea [1]. Considering etiology and demographic features, most chorea cases are associated with thyroid disorders as observed in Hashimoto's thyroiditis, females, and subjects in their mid-20s [2]. This clinical entity is rarely seen in hyperthyroidism context and the estimated incidence is < 2% among hyperthyroid individuals [3]. Here, one rare case of a 62-year-old man with unilateral hyperthyroid chorea is reported.

Case presentation

A 62-year-old man was admitted to our hospital with asymmetric involuntary movement, muscle weakness, inaccurate coordinate movement, and hypomyotonia of right limbs, which had last for 2 weeks. He also claimed one exacerbation after eating seafood. The patient had suffered from diabetes for 15 years and the blood glucose level had been controlled well. Meanwhile, he denied past illnesses of hypertension, peptic ulcer, infectious diseases, allergy, trauma, and genetic diseases. He had a history of smoking for about 40 years and 40 cigarettes per day. The patient claimed his involuntary movements of right limbs exacerbate at nightfall and his life and work had been seriously affected. (Supplement Material 1 is the video showing this patient's choretic movement). On admission, the patient's vital signs were relatively stable, but ECG showed rapid atrial fibrillation. This condition had never been discovered or diagnosed before according to the patient's own statement. MRI showed scattered patchy lesions with hazy contour, which are characterized by low signal intensity on T1 weighted sequence and high signal intensity on T2 weight sequence, around bilateral centrum ovale and corona radiate area (**Figure 1**). FLAIR imaging showed high-intensity lesions in the same region (**Figure 2**). DWI revealed patchy low signal lesions (**Figure 3**). MRA showed that A1 segment of right anterior cerebral artery was thin (**Figure 4**). SWI showed patchy low-density lesions in left cerebellar hemisphere, and phase contrast MRI showed a mixed signal. MRI also revealed some obsolete lesions in left cerebellum area. Strip-like low signal intensity on T1 weighted sequence and low signal intensity on T2 weight sequence are shown on the left side of the cerebellum. DWI also showed low

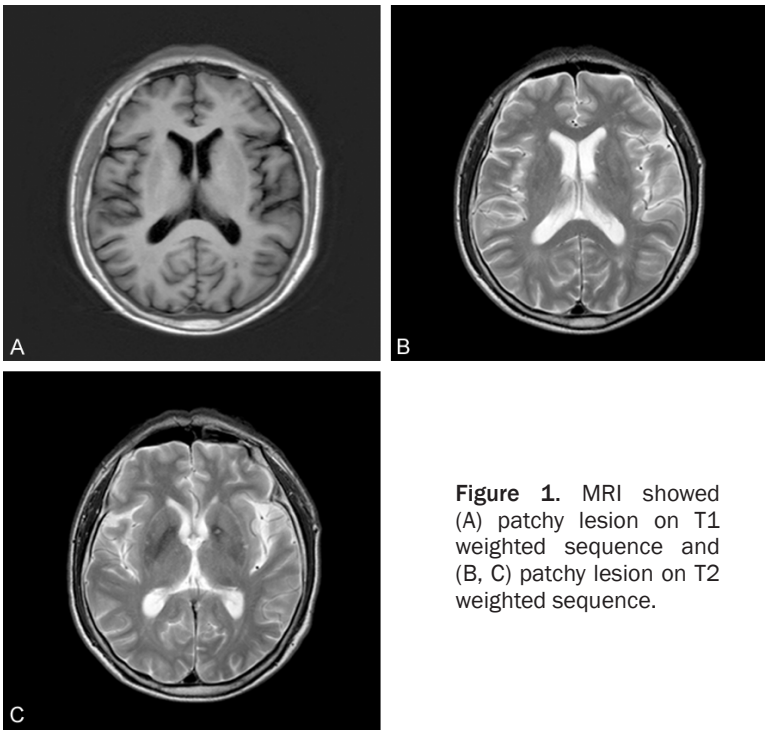

Figure 1. MRI showed (A) patchy lesion on T1 weighted sequence and (B, C) patchy lesion on T2 weighted sequence.

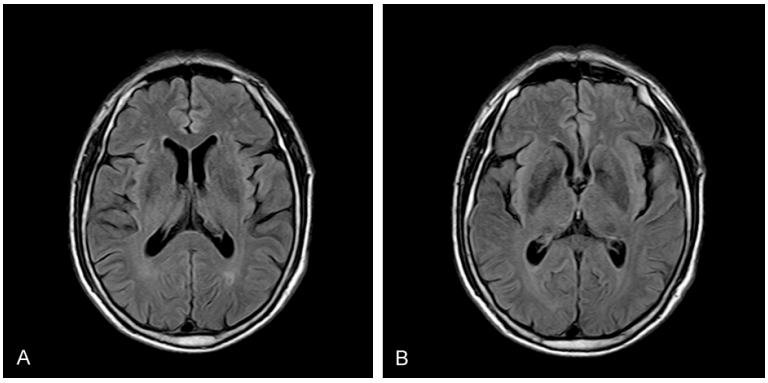

Figure 2. Flair imaging showed (A, B) high lesions with high intensity.

signal lesions in the same area (images of obsolete lesions are not shown).

FT3, FT4, T3, T4, TSH, TPO-Ab, and PTH levels were 13.94 pmol/L, 53.50 pmol/L, 3.80 nmol/L, 193.0 nmol/L, 0.00500 mIU/L, 245.7 IU/mL and 75.40 pg/mL, respectively. Other laboratory studies were all within normal limits, including blood routine examination, urine routines and urologic analysis, liver function, blood lipids, ions, renal function, coagulation routines, RF, ASO, CRP, antinuclear antibodies. Additionally, hemichorea associated with nonketotic hyperglycemia could be ruled out by adequate glucose control.

Upon admission, UCG detected tachycardia, moderate tricuspid insufficiency, and aortic insufficiency were found. Color Doppler ultrasound showed thyroid parenchymal changes.

After intramural consultations, this patient was diagnosed hyperthyroidism and arrhythmia-auricular fibrillation. Ophthalmologic examination was performed by one senior oculist, and Kayser-Fleischer rings were not detected.

Symptomatic and supportive treatments were taken to improve circulation, to nourish nerves and myocardia, to avoid increasing cardiac load. Methimazole and bisoprolol were advised to relieve and improve the symptoms.

Follow up

The patient has been followed up. He was given Thyrozol for 2 weeks after the first hospitalization, and the choretic movement improved along with the anti-thyroid therapy. Haloperidol was added to the treatment and his chorea totally disappeared. Then, 1 week later, the patient received blood tests. FT3, FT4, T3, T4, TSH, and TPO-Ab levels were 7.13 pmol/L, 12.67 pmol/L, 2.09 nmol/L, 121.25 nmol/L, 1.87 mIU/L, and 0.1 IU/mL, respectively. The patient had unauthorized withdrawal of Haloperidol for one day and he felt slight involuntary movement of the left hallux, so he went back to former therapy.

Discussion

Chorea, an abnormal involuntary movement disorder, could result from stroke, basal ganglia structural, drugs, genetic disorders, neurodegeneration, infections, autoimmune diseases, and metabolic diseases, which including thyroid diseases [4]. Hyperthyroid chorea was first

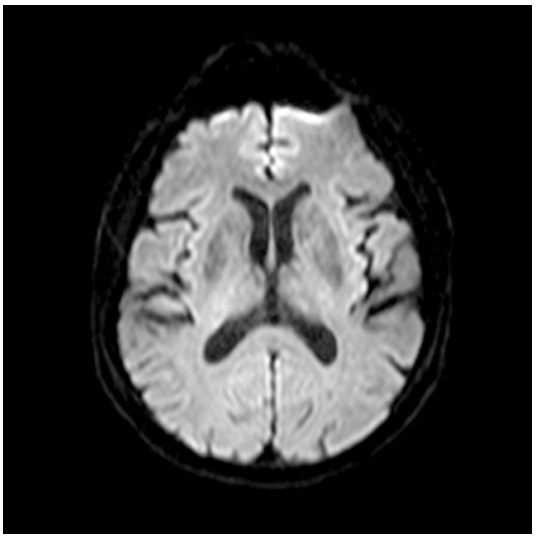

Figure 3. DWI revealed patchy low signal lesions.

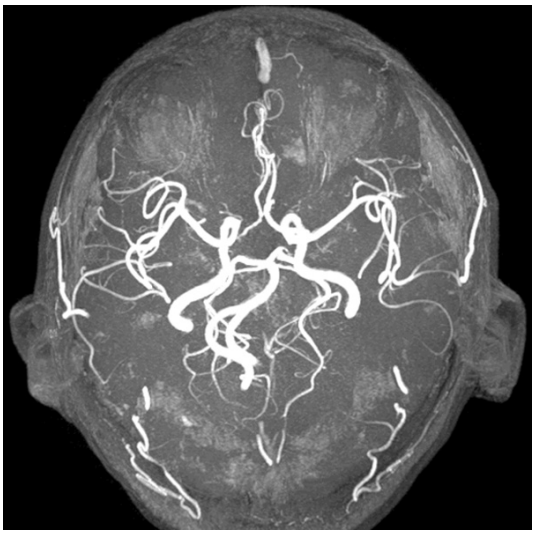

Figure 4. Thin A1 segment of right anterior cerebral artery.

recognized in 1888 and has been sporadically reported over the past century. It is still rare and poorly understood. To the best of our knowledge, hyperthyroid chorea shows a predilection for young females, and only 3 male patients have been documented by case reports so far [5]. The unique demographic features of our case may alert clinicians to be more careful about this condition in uncommon groups.

The mode of choreiform movements could be diverse. According to different reports, patients showed incapacitation at different levels--some unable to talk, chew, stand, walk and even sit [6, 7]. Bilateral ballism [3] and choreoathetosis [5-7] have been reported, and gradual development of the disease onset has been documented [8]. Furthermore, the location of chorea also varies from case to case. The extremities, trunk, face, and bucco-oral-lingual region could be affected symmetrically, asymmetrically, generally, or focally. In our observation, the patient showed asymmetric involuntary movement. By examining the patient and reviewing his medical history, the most frequent and suspicious causes of acquired chorea could be excluded, including encephalitis, Sydenham's chorea, Wilson's disease, and non-ketotic hyperglycemia status. SWI showed patchy low-density lesions in left cerebellar hemisphere. Considering the possibility of obsolete hemorrhage, we eliminated CAA by PIB-PET. Ophthalmologic examination was performed by one senior oculist, and Kayser-Fleischer rings were not detected. Combining normal ceruloplasmin and copper levels, WD was excluded. The patient denied infection. Sydenham's chorea could be ruled out by the age of disease onset, and rheumatism indicators including RF, ASO, CRP and antinuclear antibodies.

Although the mechanism behind hyperthyroid chorea remains elusive, there have already been several hypotheses trying to connect the complex basal ganglia dysfunction of chorea and effects that thyroid hormones have on the central nerve system. It was reported patients could manifest choreiform movements without pathological changes in the central nervous system [9], and another study reported one case of chorea caused by thyroxine replacement therapy [10]. These findings remind us that functional or biochemical disturbance aroused directly by hyperthyroidism may be responsible for the subsequent chorea. Klawans et al. studied effects of dopamine antagonists in both patients and animal model with hyperthyroid chorea [11, 12]. It has been suggested that hyperthyroidism leads to functional modification of dopaminergic receptors, resulting in the movement disorder. Support for this hypothesis came from the reversibility of hyperthyroid chorea with the dopamine receptor blocker haloperidol and its re-emergence on cessation of the drug [7-11]. Haloperidol could block cerebral dopaminergic receptors and improve dopamine transformation. In this case, the patient's symptom seemed to be finally

suppressed by Haloperidol, which could support the above-mentioned theory. Moreover, it's well known that thyroid hormones also have significant influences on central serotonergic and adrenergic systems. Several groups of researchers have documented rapid reductions in choreiform movements within hours of the initiation of beta-adrenergic blocker treatment, even before the clinical remission of hyperthyroidism and more importantly, without associated change in protein-bound iodine or serum thyroxine [13, 14]. In 2003, Hayashi et al. examined the effects of beta-adrenoreceptor blockade and beta-stimulation in a hyperthyroid-choreic patient. They recorded a dramatic reduction in choreic movements after giving propranolol, and they also observed increases in both involuntary muscle activities and heart rate after giving isoproterenol, the sympathetic stimulation agent [6]. Their work manifested that hyperadrenergic state could partly account for the hyperthyroid chorea phenomenon. Specifically, in this case, the unilateral manifestation may come from genetical asymmetric distribution of receptors and unilateral breakdown of blood-brain barrier, as described by other authors [15].

Disclosure of conflict of interest

None.

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