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Case Report

Fluoxetine-responsive depression in a Chinese cerebrotendinous xanthomatosis

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Abstract

Cerebrotendinous xanthomatosis (CTX) is an autosomal recessive, lipid storage disorder which is extremely rare in the Chinese population. It is characterized by progressive neurologic dysfunction and enlargement of tendon xanthomas, and is often accompanied with neuropsychiatric symptoms. Few reports are available regarding depression and antidepressant medication in CTX patients. Here, we report a Chinese case of CTX associated with fluoxetine-responsive major depression.

Keywords: Cerebrotendinous xanthomatosis; Major depression; Chinese; Fluoxetine

1. Introduction

Cerebrotendinous xanthomatosis (CTX) is a rare, autosomal recessive, lipid storage disease. Patients often exhibit progressive tendon xanthomas, juvenile cataract and neuropsychiatric symptoms due to the accumulation of cholestanol in the tissue of the Achilles tendon and central nervous system [1,2]. Some CTX cases have been described with psychotic symptoms, whereas depression is less reported [1,3,4]. Treatment with chenodeoxycholic acid (CDCA) alone or in combination with 3-hydroxy-methyglutaryl coenzyme A reductase inhibitors has been reported to halt and even reverse disease progression [3,5]. However, few reports have covered antidepressant medication in CTX patients. Here, a brief report on a Chinese case of CTX and major depression was presented to show the effectiveness of therapeutic approach with fluoxetine.

2. Case report

A 39-year-old, married, Han-nationality man was admitted to our inpatient psychiatric service with hopelessness and despondency. History was obtained from the patient and his relatives with reliability. The patient was diagnosed as having CTX by pathological examination and genetic analysis 8 months ago. Surgical excision and reconstruction of right Achilles tendon xanthomatosis were performed by the transfer of the tibialis posterior tendon, while he received ursodeoxycholic acid (UDCA) medication. Since he cannot go on with his work, he had been depressed and worried about losing his job for more than 2 months. He had poor sleep and little appetite with a severe body weight loss, He lost his interest and stopped caring for his personal hygiene and once seriously thought about committing suicide by taking sleeping pills.

According to his mother, he displayed normal development until 15 years of age when he began to show learning difficulties. He had worked in a post office for 10 years after graduating from middle school. He was introverted and showed poor sociality ability. At age 37 years, he started to

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have progressive gait instability and enlargement of the Achilles tendons. He had no seizures, no manic episode, no psychotic feature and no other remarkable medical history. There is no family history of the same disorder and neuropsychiatric illness.

The neurological examination showed bilaterally increased deep tendon reflexes and positive Babinski reflexes. He walked with a shuffling wide-based gait but showed normal motor strength of extremities. He had no cerebellar ataxia, no peripheral neuropathy and no cataracts, but had high myopia.

The mental status examination revealed depressed mood, loss of interest and decreased energy. He was especially concerned about losing his job and was very pessimistic about his future, often using such expressions as “my life is ruined” and “no work is more painful than killing me.” He admitted having recurrent suicidal ideation but denied hallucinations and delusional ideas.

Electroencephalogram analysis result was moderately abnormal with the enhancement of theta activity and short runs of irregular delta waves of 2–3 Hz. An abdominal ultrasound revealed cholecystic polypus. His blood test results — including cholesterol and triglycerides levels, and liver, kidney and thyroid function — were all in normal range. Results of X-rays of chest, bone mineral density, electrocardiogram and echocardiogram were also normal. Psychological testing revealed gross deterioration in his intellectual abilities with Wechsler Adult Intelligence Scale-Chinese Revision IQ score of 58. Magnetic resonance imaging (MRI) of the brain revealed bilaterally symmetrical hyperintensities in the dentate nucleus of cerebellar hemisphere, posterior limb of the internal capsule and brain stem, but no restriction on diffusion-weighted imaging, no space-occupying effect and no shift of cerebral midline (Fig. 1). MRI of the left ankle showed fusiform thickening and heterogeneous signals in the Achilles tendon (Fig. 2).

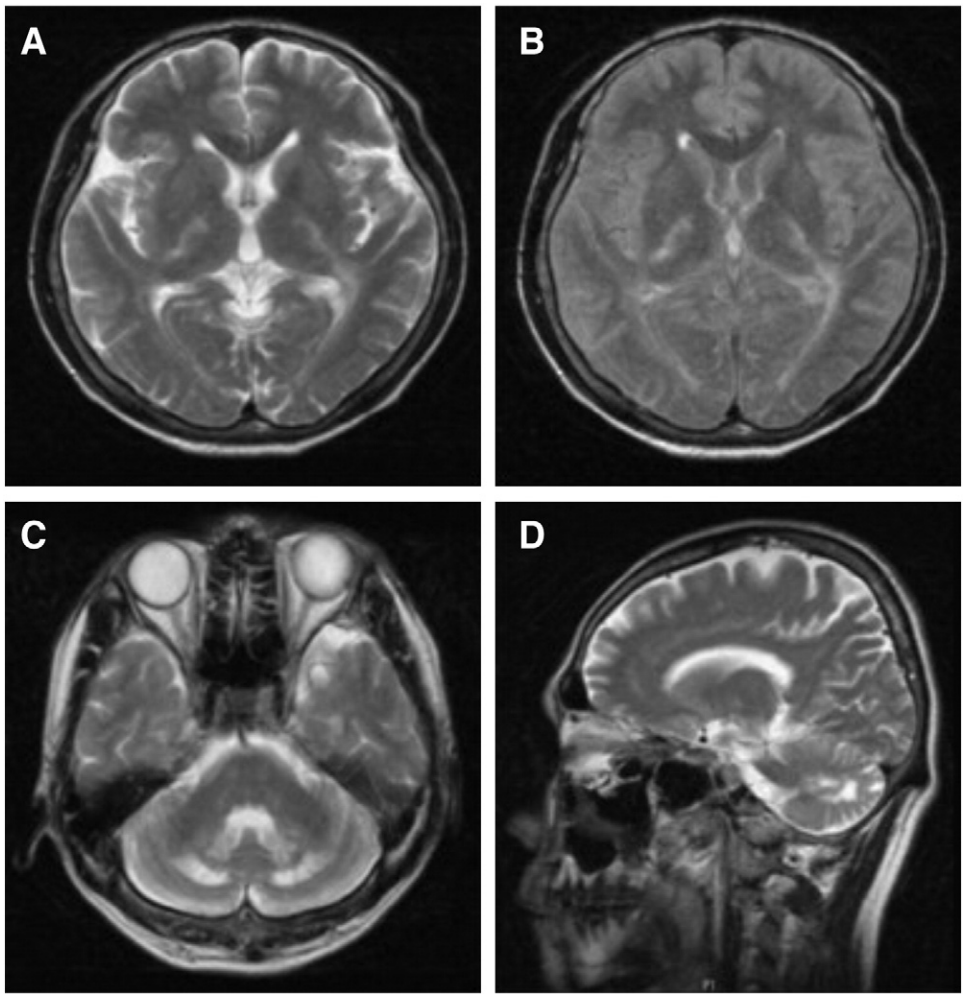

Fig. 1. Brain MRI: axial T2-weighted image (A) and Fluid Attenuated Inversion Recovery-weighted image (B) show high signal intensity symmetrically in the posterior limb of the internal capsule. Axial T2-weighted image (C) shows high signal intensity symmetrically in the dentate nucleus of cerebellar hemisphere. Midsagittal T2-weighted image (D) shows hyperintense signals in cerebellar hemisphere and mild cerebellar atrophy.