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Letter to the Editor (Case report)

Serotonin-Norepinephrine Reuptake Inhibitor (SNRI) treatment for Isaacs syndrome with depression

1. Background

Serotonin- norepinephrine- reuptake inhibitor (SNRI) has been widely used in depression and pain disorders (Goldstein et al., 2004). Duloxetine, one of the commonly prescribed SNRIs, has also been reported to be effective not only for somatic and depressive symptoms in depression but also for pain not associate with depression (Shelton et al., 2007). In fact, duloxetine has been approved by FDA for the treatment of diabetic neuropathic pain (Wong et al., 2007) and fibromyalgia (Arnold, 2007).

Pain is a devastating symptom in Isaacs syndrome, which is a rare neuromuscular disorder with continuous signaling of peripheral nerve fibers, as along with symptoms of progressive muscle stiffness and severe pain (Isaacs, 1967; NINDS, 2007). The characteristic symptoms of Isaacs syndrome include involuntary twitchings (myokymia), muscle stiffness, muscle pain, and electromyogram (EMG) of limbs often reveals myokymic discharges. (Isaacs, 1967; NINDS, 2007). To our knowledge, SNRI has not yet been reported in treating painful and depressive symptoms of Isaacs Syndrome. Here we report a patient with Isaacs syndrome and major depressive episode who responded well to duloxetine in the improvement of mood and painful symptoms.

2. Case Report

Mr. A, a 25 year-old married Taiwanese man, was admitted to the Neurology ward due to an exacerbation of Isaacs syndrome for one week. Tracing back his history, Mr. A had had continuous myokymia of lower eyelids and fingers since he was 15y/o. He first visited Neurology clinic when he was 23 year-old due to an exacerbation of muscle stiffness over his limbs and trunk, along with symptoms of facial numbness, dyspnea, pseudohypokinesia, hyperhydrosis, and myokymia. At the same time, he had involuntary movements over four limbs, trunk, eyelids, facial muscle and lips with a frequency of 5–10times/second. The EMG of his four limbs was compatible to his clinical presentation and revealed myokymic discharges at right abductor pollis brevis (APB) and left gastrocnemius (GC) muscles. He was diagnosed to have Isaacs syndrome, and had responded well to phenytoin 300 mg/d and flunarizine 75 mg/d without applying the immunoabsorption plasmapheresis therapy.

Unfortunately, his symptoms of muscle pain, myokymia, muscle stiffness, dyspnea, and pseudohypokinesia were worsened one week before his admission while receiving the regular medication therapy. In a comprehensive evaluation of psychiatric consultation, he was found to a major depressive episode (American Psychiatric Association, 1994) with symptoms of depressed mood, easy fatigue, anhedonia, hopelessness and helplessness for 1 year. Duloxetine 30 mg daily was prescribed and he experienced a complete remission in depression and somatic discomfort. To date, he is able to return to work and to carry on his daily activities for one year under duloxetine treatment.

3. Discussion

Isaacs syndrome (also known as continuous muscle fiber activity syndrome or quantal squander syndrome) is a rare familial or acquired neuromuscular disorder, which usually onset before 40 years of age. The clinical presentation of Isaacs syndrome includes progressive muscle stiffness, continuous muscle vibrating or twitching, muscle pain and cramping, sweating, and decreased reflexes. The pathophysiology of Isaacs syndrome is associated with the continuous signaling from peripheral nerve fibers, which in turn causes pain and activates muscles fibers (Isaacs, 1967; NINDS, 2007). Anticonvulsants are the standard treatment for Isaacs syndrome via alleviating muscle cramping (NINDS, 2007). In our case report here, Mr. A developed treatment resistance to the anticonvulsant, phenytoin, but had a good response to the combined SNRI therapy.

The neurotransmitters, serotonin (5HT) and norepinephrine (NE), may be important in both depression and pain (Fava, 2003). Both 5HT and NE can act as endogenous analgesics through the descending pain inhibitory pathways in the brain and spinal cord. (Fishbain et al., 1997). The theory of depression and pain sharing the common descending pathways may be applied in our case, since our patient remitted completely under the treatment of SNRI for his painful symptoms of Isaacs Syndrome and depression. However, the comorbidity of Isaacs Syndrome and depression has never been reported.

To our knowledge, this is the first report of successful treatment with SNRI for Isaacs syndrome comorbid with depression. Randomized control studies with quantified rating scales are needed to provide further support in our observation in this case.

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