

Reversible Oro-Lingual Dyskinesia Related to Lithium Intoxication

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Abstract-

Purpose: To report the first case of Taiwanese with lithium intoxication presenting as oro-lingual dyskinesia.

Case Report: A 68-year-old man had bipolar disorder with chronic lithium treatment. He had acute conscious disturbance, atrial flutter, myoclonus of limbs, and oro-lingual dyskinesia. Biochemistry study revealed elevated blood urea nitrogen, creatinine, and lithium level (3.43 Eq/L). The lithium is discontinued and he received conservational treatment. Along with reduction of serum lithium level, his involuntary movement subsided following by clear consciousness. He had no residual neurological deficit in 3 years of follow up.

Conclusion: Oro-lingual dyskinesia is a rare presentation of lithium intoxication. This case reminds us such diagnostic possibilities especially in elder patients who receive a chronic lithium therapy.

Key Words: Lithium, intoxication, oro-lingual dyskinesia, acute renal insufficiency

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INTRODUCTION

Oro-lingual dyskinesia, caused by various conditions, is one of the unusual focal dyskinesias characterized by slow semi-rhythmic jerks intermixed with dystonic movements of the mouth and tongue. Lithium is a common mood-stabilizer with high possibility of overdose side effects including diarrhea, vomiting, arrhythmia, hypotension, akathisia, coma, and seizure. Involuntary movements secondary to lithium intoxication such as tremors, ataxia, and chorea of the limbs are occasionally reported⁽¹⁾ but orolingual dyskinesia has

never been reported in Taiwan. Here, we report one patient displaying oro-lingual dyskinesia caused by lithium intoxication with eventual complete resolution after treatment.

CASE REPORT

The 68-year-old man had a history of bipolar disorder, and had been using lithium carbonate (600 mg/day) medications and sedative agents for at least 20 years. Two days before his admission, he suffered from general weakness and diarrhea. After admission, his con-

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sciousness rapidly became stuporous. One day later, he developed irregular, slow, choreaform protruding and twisting movements of his tongue along with intermittent mouth opening or pouting (Figure 1). The involuntary movement was not interfered with by outer stimuli.

Neurological examination revealed Glasgow Coma Scale of E2M4V2, intact cranial reflexes, four limbs symmetrical response to pain stimuli, and generalized increased deep tendon reflexes. Besides, positive and negative myoclonus involving four limbs were observed.

Serum laboratory findings showed elevated concen-



Figure 1. A 68-year-old man with stuporous consciousness, tongue protrusion and twisting, and intermittent mouth opening.

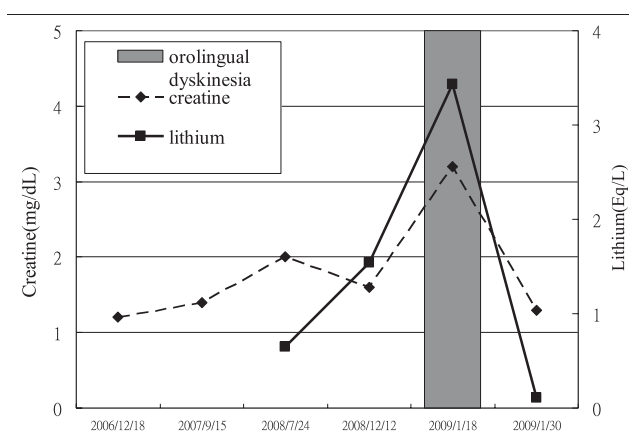


Figure 2. A graph showing the clinical course of blood creatinine and lithium concentrations indicates the occurrence of orolingual dyskinesia (hatched area) corresponding to the acute exacerbation of metabolic dysfunction.

trations of blood urea nitrogen (BUN: 51mg/dL), and creatinine (Cr: 3.2 mg/dL). In addition, a high serum lithium level 3.43 Eq/L (ref.: 0.5-1.5 mEq/L) was detected (Figure 2). Electrocardiography recorded persistent atrial flutter. Serum ammonia and sugar levels were within normal range. Electroencephalography showed generalized theta waves without epileptiform discharge. Cerebrospinal fluid analysis, brain computed tomography, and brain magnetic resonance image showed no abnormal finding.

Lithium carbonate was discontinued since detection of high serum lithium level. After supportive treatment and adequate intravenous hydration, his dyskinesia of the mouth, tongue and limbs disappeared completely 5 days later. He regained clear consciousness without an altered mentality on the 16th day after admission when his renal function (Cr: 1.3 mg/dL) and serum lithium level (0.11 mg/dL) returned to normal. In three years of follow up with medical care, he had normal neurological presentation without any involuntary movements. His serum lithium remained in the normal therapeutic range.

DISCUSSION

Lithium is a widely used medication for bipolar disorder. With its narrow therapeutic window (0.5-1.5mEq/L), lithium intoxication is frequently encountered. In this case report, the clinical symptoms of atrial flutter, hyper-reflexia, myoclonus and confusion are the typical presentation of severe lithium intoxication⁽¹⁾. His initially high serum lithium level and clinically complete resolution after normalization of serum lithium indicated a toxicity course. The diagnosis of lithium intoxication was made.

Approximately 95% of a single dose of lithium is excreted by the kidney. The half-life of lithium elimination is prolonged by renal insufficiency, advanced age, and chronic lithium use. Other predisposing factors for intoxication include overdosing, diuretics use, infection, intravascular volume depletion, or hyponatremia. In our case, we consider his diarrhea with secondary dehydration is the leading cause of acute renal insufficiency and lithium intoxication.

When serum lithium exceeds the therapeutic level, there will be symptoms such as nausea, vomiting, tremors, hyperreflexia, akathisia, ataxia, arrhythmia, or myoclonus. In severe intoxication (serum level exceed 3.5 mEq/L), confusion, coma, seizure, and even death may occur. There is no antidote for the toxicity. The management is determined by the degree of intoxication. Lithium removal with hemodialysis is required in patients with high serum levels or severe neurologic signs (seizures, stupor, coma). For mild cases, the treatment is mainly supportive with fluid resuscitation^(1,2).

Tremors are the most frequent movement disorder associated with lithium therapy⁽³⁾. There are only few case reports of oro-lingual dyskinesia secondary to lithium exposure, and tardive dyskinesia is the most common condition in these reports⁽⁴⁾. According to our knowledge and thorough research, there is no report of reversible oro-lingual dyskinesia caused by acute lithium intoxication in Taiwan. Other causes of oro-lingual dyskinesia include viral encephalitis⁽⁵⁾, anti-NMDA receptor encephalitis⁽⁶⁾, a stroke involving the brainstem^(7,8), neuroacanthocytosis⁽⁹⁾, nonketotic hyperglycaemia⁽¹⁰⁾, and hepatocerebral degeneration⁽¹¹⁾. In our case, encephalitis, central nervous system infection or structural lesion, electrolyte imbalance, hepatic encephalopathy, or blood sugar abnormality were excluded.

The reported physiological mechanisms of lithium are numerous. Lithium treats bipolar disorder by alternation of cation transport across the cell membrane in nerves and influences serotonin and/or norepinephrine reuptake⁽¹²⁾. How lithium cause oro-lingual dyskinesia is not clearly known but a few pathogenic mechanisms have been presumed. According to studies on tardive dyskinesia, the possible pathophysiology of oro-lingual dyskinesia after chronic lithium exposure includes GABA insufficiency⁽¹³⁾, reduced dopamine in the striatum⁽¹⁴⁾, and dopamine receptor hypersensitivity⁽¹³⁾. For acute lithium-induced extrapyramidal symptoms, anticholinesterase effect is speculated⁽¹⁵⁾. In one rat study, injections of the cholinergic agonist into the globus pallidus elicited tongue protrusions⁽¹⁶⁾. In our case, the symptoms of atrial flutter and myoclonus may also be secondary to increased cholinergic activity.

Lithium intoxication is not uncommon, especially in patients of advanced age or chronic lithium exposure. Tremors are the most frequent movement disorder associated with lithium therapy. This is the first report of lithium induced reversible oro-lingual dyskinesia in Taiwan. This case highlights the diagnostic possibilities of lithium intoxication in patients with acute oro-lingual dyskinesia.

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